Fasting intolerance and recurrent hypoglycemia: Ponder for Sheehan's

Sir,

Sheehan's syndrome is a clinical entity, which occurs because of pituitary apoplexy after severe postpartum hemorrhage (PPH). This syndrome is uncommon to be picked up in the early postpartum period. We describe a case of Sheehan's syndrome, who presented because of recurrent hypoglycemia with shock 3 months after PPH in her last delivery. The presentation of Sheehan's syndrome with recurrent hypoglycemia in so early period after PPH is rarely reported.

A 22-year-old female came to the hospital because of the history of recurrent episodes of perspiration and irrelevant talking, followed by unconsciousness since 3 months after PPH. Initially, she had fasting intolerance followed by these episodes which were progressive over a period of 3 months. She gave the history of regaining consciousness after infusion of fluids in the peripheral hospitals. There was no history of chest pain, fever, headache, vomiting, trauma, or seizures. She had no history of chronic illness or addictions. She delivered her last baby at home 4 months back when she was gravida 2, parity 1 during which severe PPH occurred, for which she was hospitalized, and 2 units of packed cells were transfused. She did not breast feed the baby. On examination, she was semiconscious, cool, and clammy. The systolic blood pressure was 60 mmHg, pulse rate was 118 beats/min regular, respiratory rate was 18 breaths/min, temperature was 98.5°F by axilla, and blood sugar was 23 mg/dL. She regained consciousness after bolus intravenous infusion of dextrose solution. There was dryness of skin, conjunctival pallor, facial puffiness, nonpitting pedal edema, delayed ankle jerk, and slowness of speech. Pubic and axillary hairs were scanty. The cardiac, respiratory, gastrointestinal, and nervous system examination did not reveal any abnormality.

Hematology revealed hemoglobin 8.5 g/dL with normal leukocyte and platelets count. The peripheral blood film showed microcytic hypochromic red cells. The iron profile was consistent with the diagnosis of iron deficiency anemia. Liver function tests, renal function tests, and electrolyte were within normal limit. The hormone profile showed a deficiency of anterior pituitary hormones [Table 1]. The contrast enhanced magnetic resonance imaging of pituitary fossa showed empty sella [Figure 1a and b]. The deficiency of anterior pituitary hormones, brain imaging, clinical history, and physical examination were consistent with the diagnosis of panhypopituitarism because of Sheehan's syndrome. She was treated with intravenous fluids, glucocorticoid replacement, and then thyroxin was added. At 1-month follow-up, the patient was apparently asymptomatic.

Sheehan's syndrome is one of the leading causes of hypopituitarism in young females in the underdeveloped or developing countries. This occurs due to ischemic pituitary necrosis after severe PPH. The exact mechanism is still unclear. The pituitary hypoperfusion and necrosis are considered because of hypotension and anterior pituitary arterial vasospasm, thrombosis, or vascular compression following severe PPH.[1,2] The onset of signs and symptoms of hypopituitarism depends on the degree of anterior pituitary hormone deficiency and usually does not occur till the 75% of pituitary damage.[2] The clinical spectrum of Sheehan's syndrome is wide and varies from nonspecific symptoms such as weakness, fatigue, and anemia to severe secondary adrenal failure and hypothyroidism resulting in coma and death. Among the common clinical symptoms, absence of lactation is the first to appear after the delivery. Other symptoms of the syndrome are amenorrhea, loss

Table 1: Hormone profile of the patient at the time of diagnosis

Hormone	Value	Reference value
Serum cortisol (8.00 a.m.)	11.04	138-690 nmol/L
Serum ACTH	2.62	1.3-16.7 pmol/L
Serum FT4	<5.15	09-16 pmol/L
Serum FT3	<1.54	3.7-6.5 pmol/L
Serum TSH	4.4	0.34-4.25 mIU/L
FSH	7.4	Postmenopausal 18-153 IU/L
LH	1.4	Postmenopausal 16-64 IU/L
E2	45	<121 pmol/L
Serum prolactin	1.83	40-530 mIU/L
GH	0.05	0-5 μg/L
Serum insulin	1.32	14.35-143.5 pmol/L
Serum C-peptide	<0.02	0.27-1.19 nmol/L

ACTH: Adrenocorticotropic hormone, FT: Free thyroxin, TSH: Thyroid stimulating hormone, FSH: Follicular stimulating hormone, LH: Luteinizing hormone, E2: Estradiol, PRL: Prolactin, GH: Growth hormone

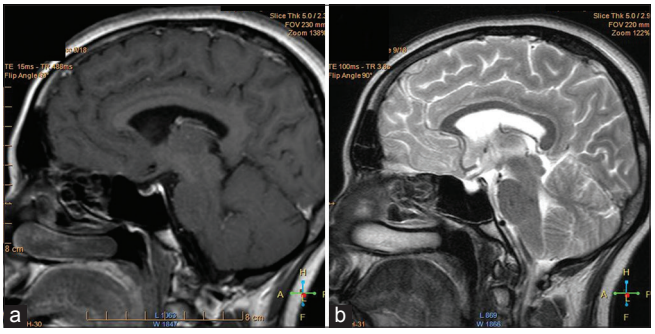

Figure 1: (a and b) The contrast enhanced magnetic resonance imaging of pituitary fossa showing empty sella

of libido, loss of pubic and axillary hair, hypoglycemia, and symptoms related to central hypothyroidism.[2,3] The diagnosis of Sheehan's syndrome immediately after severe PPH is reported in few cases. Majority of patients remain undiagnosed for decades. Sheehan's syndrome rarely present with recurrent hypoglycemia, which occurs because of adrenal insufficiency and lack of counter-regulatory mechanisms of hypoglycemia.[4-7]

Our patient presented because of recurrent hypoglycemia and she also had the symptoms related to central hypothyroidism, lactation failure, and anemia. The possibility of lymphocytic hypophysitis, which is a rare autoimmune entity common in pregnancy, was excluded because of the absence of clinical sign and symptom of the disease, presence of typical obstetric history, and imaging studies. The cause for recurrent hypoglycemia in our patient was acute adrenal insufficiency with absent counter-regulatory mechanisms. An isolated adrenal insufficiency is rarely severe enough to cause hypoglycemia. There was a lack of counter-regulatory mechanisms because of steroids and growth hormone deficiency, secondary to hypopituitarism, which precipitated hypoglycemia. The cortisol deficiency depletes glycogen store, decreases gluconeogenesis, and blunts epinephrine response in symptomatic hypoglycemia. The very low level of serum insulin and c-peptide along with decreased anterior pituitary hormones is explained by decreased secretion of insulin in response to hypoglycemia, to prevent further deterioration of hypoglycemic attack. The electrolytes were normal in secondary hypoaldosteronism as aldosterone is under the control of rennin angiotensin system also.

To conclude, patient presenting with "fasting intolerance and recurrent hypoglycemia" after PPH and having clinical picture suggestive of hypothyroidism should be evaluated for Sheehan's syndrome and hormone replacement should be considered at the earliest.

Financial support of sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

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Access this article online	
Quick Response Code:	Website: www.ijem.in
	DOI: 10.4103/2230-8210.190593

Cite this article as: Pahadiya HR, Lakhotia M, Gandhi R, Yadav RS, Madan S. Fasting intolerance and recurrent hypoglycemia: Ponder for Sheehan's. Indian J Endocr Metab 2016;20:739-40.