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Case Report

Recurrent Hypoglycaemia: an Uncommon Presentation in Sheehan Syndrome

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ABSTRACT

Sheehan's syndrome is a rare but potentially serious postpartum complication. Though most common and early symptoms are lactation failure and amenorrhea, some cases might be relatively asymptomatic which are diagnosed in later years when features of hypothyroidism and adrenal insufficiency predominate. Recurrent hypoglycaemia, due to adrenal insufficiency, though described is a rare complication of Sheehan syndrome. Here we report a case of Sheehan syndrome which presented with recurrent episodes of hypoglycaemia,

Key words: *Sheehan's syndrome, hypoglycaemia, hypothyroidism, adrenal insufficiency*

Malign Servikal Lenf Nodlarinin Ayiriminda ve Yönetiminde Ultrasonografinin Rolü

ÖZET

Sheehan sendromu nadir fakat ciddi bir postpartum komplikasyondur. En sık ve erken belirtileri arasında laktasyon yetmezliği ve amenoredir. Bazı durumlarda Sheehan sendromu hipotiroidizm ve adrenal yetmezliği özellikleri baskın olduğunda daha sonraki yıllarda tanı konulan asemptomatik olabilir. Adrenal yetersizlik nedeni ile tekrarlayan hipoglisemi Sheehan sendromunun nadir görülen bir komplikasyonudur. Burada, tekrarlayan hipoglisemi atakları ile sunulan bir Sheehan sendromu olgusu bildirildi.

Anahtar kelimeler: *Sheehan sendromu, hipoglisemi, hipotroidizm, adrenal yetersizlik*

INTRODUCTION

Sheehan's syndrome is a rare but potentially serious postpartum complication. First described by Sheehan in 1937; it is the ischemic necrosis of the pituitary gland secondary to shock due to obstetric hemorrhage (1). It is a well-known cause of hypopituitarism secondary to pituitary apoplexy. Most common presenting symptoms of Sheehan's syndrome are lactational failure, prolonged amenorrhea or oligomenorrhea after delivery (2). Rarely, a woman with Sheehan syndrome might be relatively asymptomatic, and the diagnosis is not made until years later, with features of hypothyroidism and adrenal insufficiency (2). Recurrent hypoglycaemia and hyponatremia though described in literature are less known complication of Sheehan's syndrome with only few case reports (3). We report a case in which patient who had recurrent episodes of altered sensorium due to hypoglycaemia on detailed history and investigations was diagnosed as sheehan syndrome.

CASE

A 48-year-old female patient presented to the outpatient department of our hospital in January 2014 with history of multiple episodes of sudden loss of consciousness for past 15 years. Patient also complained of generalized swelling of body and weakness with inability to do household work since past 5 years. On detailed history it was revealed that the woman had multiple admissions in past for episodes of unconsciousness during which her blood glucose was recorded below 40 mg with frequency of episodes around 2 per year. During all these episodes patient regained consciousness on administration of intravenous glucose. There was no history of any other drug intake.

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Patient 17 years back had intrauterine death of seven month foetus following which she developed post-partum haemorrhage and had to be transfused with multiple units of blood. She has amenorrhea since then for which she did not seek medical attention.

On examination she was conscious, oriented to time, place and person. Her pulse rate was 70 per min, regular, low volume and blood pressure was 90/60 mm Hg with a pale and rough skin. She had hoarseness of voice, dry skin, thinning of hair, facial puffiness, non-pitting pedal oedema. Her systemic examination was unremarkable. Patient had weakness of proximal muscles in both upper and lower limbs with power 4/5. Distal muscle power was essentially normal. Her ankle reflex was present but delayed. Sensory system examination was essentially normal. Initial investigations showed a anemia with normal indices and reticulocyte count. Her ESR, renal function, liver function tests and serum electrolytes were normal. Her thyroid profile showed low free triiodothyronine (FT3) and free thyroxine (FT4) with normal thyroid stimulating hormone which pointed towards the diagnosis of central hypothyroidism. Her fasting 0800 hrs serum cortisol levels were found to be 1.23 µg/ml which was very low.

Elaborate endocrinological investigations were done which showed LH, FSH deficiency and GH, ACTH insufficiency further confirming our diagnosis of central hypopituitarism (Table 1). Her vitamin D levels and DEXA score were normal. Patient's plasma insulin and C-peptide levels were normal ruling out the possibility of insulinoma as reason behind hypoglycaemia. Her ECG, chest X-ray PA and abdominal ultrasonography showed no abnormality with normal adrenals. A magnetic resonance imaging of the pituitary gland showed an "empty sella" appearance with hypoplastic pituitary flattened against the sellar floor with normal bright spot of posterior pituitary (Figure 1). Patient was treated with hydrocortisone 15 mg in morning and 5 mg in evening. L-thyroxine in a dose of 0.075 mg was started, to be titrated on basis of subsequent T4 levels. Somatotrophin was started in a dose of 1 mg qd. On regular follow up patient improved symptomatically with no episodes of hypoglycaemia. She has been further advised to increase the dose of hydrocortisone during any stressful event.

DISCUSSION

Adenohypophyseal ischemic necrosis following hypo perfusion is the most common cause of adenohypophysal insufficiency.Vasospasm, thrombosis, and vascular compression of the hypophyseal arteries could be the possible causes of Sheehan's syndrome. The classical condition associated is postpartum hemorrhage with cardiovascular collapse (4). The most common antecedent obstetric events described with this syndrome includes, obstetric haemorrhage (82%), shock (47%), blood transfusion (43%), and peripartum hysterectomy (7%) (5).

The diagnosis can be made reliably in the presence of lactational failure, prolonged amenorrhea. However, other signs of adenohypophysal insufficiency like hypothyroidism and adrenal failure are often delayed and subtle leading to the diagnosis being missed. In some cases the pituitary necrosis is only partial and the syndrome can present in atypical and incomplete forms further complicating the diagnosis (6). Recurrent hypoglycaemia and hyponatremia are rare presentation of sheehan syndrome. Although described earlier, there are only few case reports of sheehan syndrome presenting as recurrent hypoglycaemia. The female in our case presented with symptoms of recurrent loss of consciousness which were documented due to recurrent hypoglycaemia. Such recurrent episodes of altered sensorium 15 years after puerperal haemorrhage, is a rare presentation of Sheehan's syndrome.

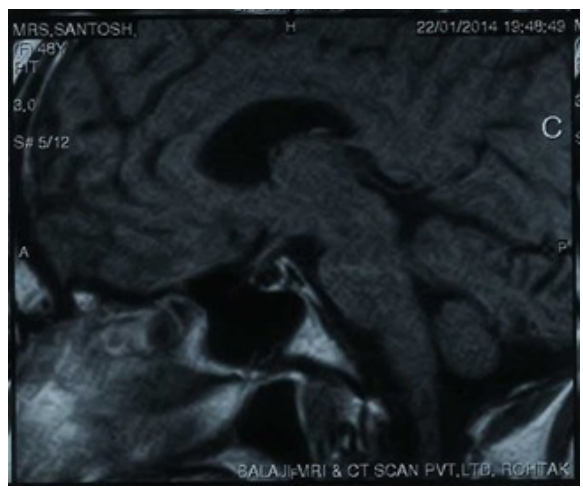

***Figure 1**. MRI showing empty sella" appearance with hypoplastic pituitary flattened against the sellar floor with normal bright spot of posterior pituatory*

***Table 1**. Investigational Profile of Patient.*

Parameter	*Patient*	*Normal*
Haemoglobin	*9.3*	*12-16 gm/dl*
TLC	*7000*	*4000-11000/cumm*
APC	*1.8*	*1.5-4.5 lac/cumm*
PBF	*N/N*	
Serum Na+/K+:	*136/4.8*	
HbA1C:	*5.4%*	
Thyroid Profile:		
FT3	*1.02*	*1.71-3.71 pg/ml*
FT4	*0.08*	*0.70-1.48 pg/ml*
TSH	*0.79*	*0.35-4.94 mIU/ml*
Serum Cortisol (0800hrs)	*1.23*	*6.2-19.4 µg/ml*
FSH	*0.95*	*26.72-133.41 mIU/ml*
LH	*0.25*	*10.39-64.57 mIU/ml*
GH (Insulin tolerance test)		
0 min	*<0.03*	*Normal Response is GH*
30 min	*<0.04*	*increase to >3 µg/ml*
60 min	*<0.04*	
ACTH (Insulin tolerance test)		*Normal Response is Cortisol should*
Cortisol at	*2.43*	*increase by >7 µg/ml or*
0 min	*2.88*	*to >20 µg/ml*
30 min	*2.90*	
60 min		
Plasma Insulin levels(BG <55mg%)	*2.57*	*2.6-24.9 µU/ml*
Vitamin D	*29.8*	*15-30 ng/ml*
DEXA Scan		
T Score	*0.9*	
Z Score	*0.8*	

The ability of human body to tolerate fasting is dependent on glycogen stores and gluconeogenesis. Deficiency of cortisol causes anorexia and weight loss which leads to depletion of body glycogen stores.This increases the body reliance for glucose production on gluconeogenesis pathway. The low-level of gluconeogenesis precursors (like gluconeogenic amino acids and fatty acids), due to cortisol deficiency and the glycogen depletion results in impaired ability to tolerate fasting. Moreover, the growth hormone deficiency also contributes to the hypoglycemia (7). This patient was found to have low levels of cortisol and growth hormone, which contributed to impaired fasting tolerance and recurrent episodes of hypoglycaemia. This improved after treatment with hydrocortisone and growth (8). These findings were similar to those by Bala M et al and Selcuk Yaylaci et al who reported similar cases of female patients presenting with hypoglycemia and hyponatremia long after delivery who were later diagnosed as Sheehan syndrome (9,10). The patients in both previous case reports had cortisol and growth hormone deficiency which was also present in our patient, highlighting their importance in hypoglycemia.

To conclude, in a woman having recurrent episodes of hypoglycaemia with background history of previous pregnancy with post-partum haemorrhage and secondary amenorrhea, Sheehan's syndrome though overlooked as uncommon is one of the differentials to be sought for.

REFERENCES

1. *Sheehan HL. Post-partum necrosis of the anterior pituitary. J Pathol Bacteriol 1937;45:189.*
2. *Schrager S, Sabo L. "Sheehan syndrome: a rare complication of postpartum hemorrhage". J Am Board Fam Pract 2001;14(5):389-91.*
3. *Dosi RV, Saraiya S, Patell RD, Sonune NN, Sheth SD. An unusual association of sheehan's syndrome with pancytopenia. J Case Rep 2012;2:21-3.*
4. *Assan R.Urgences métaboliques et endocriniennes In : Kleinknecht D,éd. Principes de réanimation médicales, 3e éd.Paris : Flammarion médecine sciences, 1984 :292- 326*
5. *Gei-Guardia O, Soto-Herrera E, Gei-Brealey A, Chen-Ku CH. Sheehan syndrome in Costa Rica: Clinical experience with 60 cases. Endocr Pract 2011;17:337-44.*
6. *Shivaprasad C. Sheehan's syndrome: Newer advances. Indian J Endocrinol Metab 2011;15:S203-7.*
7. *Davis SN, Cryer PE. Hypoglycemia. In: Longo DL, Kasper MD, Jameson JL, Fauci AS, Hauser SL, Localzo J, editors. Harrison's Principles of Internal Medicine. 18 th ed., Vol. 2. New York: McGraw Hill; 2012. p. 3003-9.*
8. *Singhania P, Singh S, Banerjee R, Singhania B, Banerjee I, Maitra S. Hyponatremia - A rare and emergency presentation of Sheehans Syndrome. Pak J Med Sci 2010;26:713-5.*
9. *Bala M, Brünnler T, Guralnik V, Schölmerich J, Schäffler A. A 36-year old female patient presenting with hypoglycemic coma. Internist (Berl) 2009;50:606-11.*
10. *Yaylaci S, Demir MV, Aytürk S, Tamer A. Sheehan's syndrome: Presented with hyponatremia and hypoglycemia after 14 years from delivery. Indian J Endocrinol Metabol 2012;16(2):322-3*