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Case Report

Psychosis as a Rare Late Manifestation of Sheehan's Syndrome

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Abstract

Sheehan's syndrome is defined by varying degrees of anterior pituitary hormones deficiency due to postpartum ischemia of the pituitary gland after massive bleeding. It can present during postpartum or several months or years following delivery. Endocrinologic manifestations of hypopituitarism reveal the deficiencies of specific hormones secreted from pituitary gland including hypoadrenalism, hypothyroidism and hypogonadism. Psychiatric manifestations of Sheehan syndrome has rarely been studied systematically and only few case reports are exist in this field. In this article, I presented a case who diagnosed early with Sheehan syndrome and maintained well on medications for 18 years, but recently she developed psychosis. At the end I discussed the challenging in making the diagnosis, the etiopathological basis and the treatment.

Keywords: *Sheehan Syndrome; Hypopituitarism; Psychosis; Hormonal Replacement Therapy; Antidepressants*

Introduction

Sheehan's syndrome is defined by varying degrees of anterior pituitary hormones deficiency due to postpartum ischemic necrosis of the pituitary gland after massive bleeding. was first described by an English pathologist, Harold Leeming in 1937 [1]. In an international data base containing 1034 patients with growth hormone deficiency, Sheehan's syndrome was found to be the cause in 3.1% of the cases [2]. And in a retrospective nationwide analysis in Iceland, the prevalence of Sheehan's syndrome in 2009 was estimated to be 5.1 per 100,000 women [3]. In another epidemiological study in Kashmir valley estimated the prevalence of Sheehan's syndrome to be 2.5 - 4% among parous women older than 20 years [4].

The underlying process leading to Sheehan's syndrome is the infarction of the physiologically enlarged anterior pituitary lobe (due to hyperplasia of prolactin secreting cells from elevated estrogen secretion) and secondary to the compression of the blood vessels supplying the gland by the enlarged gland itself or due to grossly decreased blood supply during intra-partum or postpartum events. Although other etiologies such as vasospasm, autoimmunity, small sella size, and disseminated intravascular coagulation may also have role in the development of Sheehan's syndrome, none has been conclusively proven [5].

It can present during postpartum period or several months or years following delivery. The mean duration between postpartum bleeding and the subsequent development of symptoms varies from 1 to 33 years [6].

Most common initial symptoms of Sheehan's syndrome are agalactorrhea and/or amenorrhea. Endocrinologic manifestations of hypopituritisim reveal the deficiencies of specific hormones secreted from pituitary gland including hypoadrencortico-tropinemia, hypothyroidism and hypogonadism. Deficiency in corticotrophin is characterized by a decrease in adrenal androgens and production of cortisol. Acute loss of adrenal function is a medical emergency and may lead to hypotension and death if not treated. Signs and

symptoms of corticotrophin deficiency include myalgias, arthralgias, fatigue, headache, weight loss, anorexia, nausea, vomiting, abdominal pain, altered mental state or altered consciousness, dry wrinkled skin, decreased axillary and pubic hair, anemia of chronic disease. And impaired gluconeogenesis. Gonadotropin deficiency causes amenorrhea, hot flushes and decreased libido. Growth hormone deficiency causes many vague symptoms including fatigue and decreased muscle mass. Tiredness, intolerance to cold, constipation, weight gain, hair loss, slowed heart rate and thinking are manifestations of hypothyroidism [7-9].

Uncommonly, it can present as an emergency condition with circulatory collapse, severe hyponatremia, diabetes insipidus, hypoglycemia, congestive cardiac failure or psychosis [10]. There are very few case reports of Sheehan's syndrome presenting as psychosis or depression [11-13]. In this article I present a case who referred to psychiatry due to psychosis as a rare sequel of Sheehan's syndrome.

Diagnosis of the syndrome is based on the clinical features of associated hormone deficiency, a suggestive obstetric history, laboratory finding of decreased hormone levels and related radiological features. Its treatment requires lifelong replacement of the deficient hormones.

Case Report

A 46 years old Saudi female, married and mother of 3 living daughters, works as a teacher, who was referred from internal medicine to psychiatry department with complaints of changes in her behavior and not being in her usual state, she doesn't want to sit and refuses to take medications. Together with talkativeness, talking to herself and sleeplessness, all were of recent onset.

18 years ago, during the delivery of her last child, patient had sever postpartum hemorrhage in the hospital that necessitate blood transfusion. Following this, patient had developed amenorrhea and lactation failure along with other symptoms. At that time diagnosis of Sheehan's syndrome had been established and patient described prednisolone 5 mg daily and levothyroxine 100 mcg daily.

In the recent few months patient quitted the medications by herself. As a result she gradually started to develop mild to moderate epigastric pain, diarrhea up to 8 times a day, nausea and vomiting up to 5 times a day, loss of appetite, fatigability, lethargy and muscle cramps. So her family brought her to the emergency where she has been stabilized and then shifted to the medical ward.

There was no past or family history of psychiatric illnesses and she was neither a smoker and had never abused illicit drugs. On general examination patient was afebrile and vitally stable, pale and her breasts were atrophied. The systemic examinations were normal. On mental status examination patient looks around her stated age, wearing clean clothes with good self hygiene. She was confused and disoriented. She exhibited good eye contact but was uncooperative and refuses to answer some questions. There were pressured speech and flight of ideas. Visual hallucinations was there. No suicidal or homicidal thoughts. She had no insight and poor judgment. Memory and cognitive functions were intact.

Her laboratory investigations including renal functions, liver functions, as well as workup for inflammatory and infectious conditions didn't reveal any abnormalities. Her hormonal profile (Table 1) revealed decreased cortisone, free T4, prolactin, FSH, LH and normal TSH.

Hormone	Patient's value	Normal value
Cortisone	96.35	171.00 - 536.00 nmol/L
Free T4	1.090	12.000 - 22.000 pmol/L
TSH	1.370	0.250 - 5.000 mIU/L
prolactin	35	102 - 496 mIU/L
FSH	0.527	
LH	0.100	
Estradiol	18.35	

Table 1: *Patient's hormonal profile.*

In the emergency unite, patient was started on hydrocortisone 100 mg and thyroxin 100 mcg. When she shifted to medical ward she was maintained on prednisolone 5 mg/day to be continued along with thyroxin 100 mcg/day. For psychotic symptoms she was managed with oral olanzapine 5 mg/day at bed time. Her psychotic symptoms was showing significant improvement, and after few days it resolved completely. Due to prolonged QTc we have chosen not to continue olanzapine and to discharge the patient with only prednisolone 5 mg/day along with thyroxin 100 mcg/day. During the follow up visit, patient was doing well and free of any psychotic or mood symptoms.

Discussion

In this article, I presented a 46 years old female patient who diagnosed with Sheehan syndrome after she had sever postpartum hemorrhage during the delivery of her last child 18 years back. Her symptoms of hormone deficiencies started early, her laboratory investigations along with suggestive obstetric history all strongly supported the diagnosis of Sheehan syndrome. So she had been diagnosed early and managed with hormone replacement therapy and maintained well on follow ups for the whole last 18 years. However, as the damage of pituitary gland is irreversible the hormonal replacement therapy should be continued for life and discontinuing it may causes serious and life threatening complications. Psychosis is one of the rarest but serious complications that can occur as a result of sudden drop of these hormones, and that what most likely happened to our patient in the last few months when she discontinued her medications.

Psychiatric manifestations of Sheehan syndrome has rarely been studied systematically and only case reports are exist in this field. Moreover to the few Sheehan syndrome linked psychosis in the literature, there are even rarer cases of late onset psychosis in Sheehan syndrome [14]. They might be presenting symptoms or might appear later in the course of the disease. They might be manifestations of the disease itself, or may be generated as complications of treatment with corticosteroid. It can be another independent diagnosis also. Thus it was difficult to establish a diagnosis.

The etiopathological basis of such associations is poorly understood, might be resulting from effect of sudden drop of several hormones on brain [11-13,15,16]. The pituitary tissue necrosis might also initiate an autoimmune process which may explain the delayed onset of clinical manifestations [17]. However, it could happened any time and it shouldn't be missed or neglected.

It has been reported that the treatment with hormonal replacement therapy only will result in complete recovery from Sheehan's syndrome related psychosis after attaining euthyroid and eucortisolemic state without using any antipsychotics [11]. However, the antipsychotics may be necessary as an adjunct therapy in the initial stages [18].

Conclusion

Psychosis in patients with Sheehan's syndrome is uncommon. However, a high index of suspicion is needed. Early detection and treatment of the psychiatric manifestation with antipsychotic as well as lifelong management of the Sheehan Syndrome with hormone replacement is absolutely necessary to prevent morbidity and mortality. Keen monitoring of the treatment and the course of the illness is also of outmost importance to detect any new symptoms and therefore, to take appropriate measures.

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Conflict of Interest

None declared.

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