

Psychotic Disorder and Sheehan's Syndrome: Etiology or Comorbidity?: A Case Report



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SUMMARY

Sheehan's Syndrome, also called postpartum hypopituitarism, is a syndrome characterized by heavy bleeding, during or after delivery, and necrosis of the pituitary gland due to hypovolemic shock. Sheehan syndrome presents with agalactorrhea, amenorrhea, hypothyroidism and hypoglycemia, in addition to psychiatric manifestations, such as psychosis. In this study, we report a case of Sheehan's syndrome that presented with a psychotic disorder. The patient was a 44 year-old, female patient, married. She was admitted for withdrawal, irritability, insomnia, hearing voices telling her to harm herself, do evil to others, and thoughts about that her husband was in an adulterous relationship. She was diagnosed as having a psychotic disorder and treated with olanzapine 20 mg/day. She had hypopituitarism symptoms, thus hormone tests and a cranial MRI were performed. Sheehan's syndrome was diagnosed and prednisolone and thyroxine were added to the treatment. Her symptoms disappeared one month later. Olanzapine was stopped after 4 months and her treatment continued with prednisolone and thyroxine. Studies concerning the etiology of psychotic symptoms refer to endocrine and autoimmune systems. In this study, we discuss a case that in which the diagnosis was a psychotic disorder and Sheehan's Syndrome, diagnosed 24 years later, and the etiological aspect, including the follow-up period and treatment.

Key words: Sheehan's Syndrome, postpartum hypopituitarism, psychosis

INTRODUCTION

Sheehan syndrome was first described by an English pathologist, Harold Leeming in 1937. The syndrome is also known as postpartum hypopituitarism. Sheehan syndrome is characterized by the presence of a hypophysial necrosis due to severe hemorrhage and hypovolemic shock (Sheehan 1937). Initial symptoms usually involve agalactorrhea and amenorrhea. Some patients may develop hypothyroidism and adrenal failure many years after a long lasting asymptomatic period. Sheehan syndrome may rarely present with psychotic disorders (Hanna 1970, Bahemuka 1981, Gupta 1995). In this article we present a patient who presented with a psychotic syndrome and accompanying Sheehan syndrome.

CASE

A 44 year old female, illiterate and working as a homemaker, presented with symptoms of autistic features, unwillingness to speak, nervousness, insomnia, and voice hallucinations.

She also said that her daughter was defaming her throughout the neighborhood and her husband was seducing her in the presence of other women. Initial symptoms began 10 months ago by with development of voice hallucinations with instructions of self-harm, excessive anxiety, and autistic features. At this time she presented to a psychiatry clinic and was started on olanzapine and citalopram with a preliminary diagnosis of psychotic depression. After one month of treatment, the patient had partially recovered and stopped taking her medications. She was free of symptoms for a six month period and did not go to her follow-up appointments. By the end of this period, the same symptoms restarted and led her to be hospitalized in our clinic for further evaluation.

The patient's obstetric medical history includes her first parity at 18 years old without any complications, a second parity at age 20, with a complication of severe hemorrhage. During the postpartum period she suffered from agalactorrhea, amenorrhea and infertility. Throughout for 24 years postpartum she had the symptoms of amenorrhea and fatigue. Just after her second parity she also had these symptoms, but a diagnosis

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could not be determined at that time. Her hypothyroid symptoms including intolerance to cold, constipation, drying of dermis and hair loss, which presented in a progressive fashion during last 10 years. She denied both a history of substance abuse and family history of any psychiatric disease.

Vital findings were stable, with general wellness in the physical examination. The only pathological finding observed was dry skin. In her psychological evaluation; she appeared older than her stated age, though her appearance was appropriate for her sociocultural situation and she was well groomed. She exhibited poor eye contact during the examination with the physician. She was oriented and cooperative with a clear attention to person and time. She was with normal intelligence and memory. Her affect was anxious with normal perceptions. Her speech was goal orientated. Her opinions harbored paranoid delusions and she described self-insulting voice hallucinations. She also presented with psychomotor retardation. Neurologic examination did not reveal any pathology.

The patient was admitted to the hospital with a preliminary diagnosis of psychotic disorder and she was planned to undergo further examinations to excluded any other medical condition. Since the patient was exhibiting symptoms compatible with Sheehan syndrome, hormone levels and hypophyseal MRI examinations were performed. Hormone levels were as follows: fT4:<0.25, fT3:2.01, TSH:0.5, FSH:4.11, LH:1.02, GH: <0.05, IGF-1:25, prolactin: 1.74, oestradiol: <20, progesterone:<0.08, cortisol: <0.4. When we bring together all these items regarding hormone levels, presence of severe hemorrhage with delivery, agalactorrhea and amenorrhea history, empty sella image in MRI, we diagnosed the patient with Sheehan syndrome. The treatment, as recommended by endocrinology was levothyroxine sodium 150 mcg/day plus prednisolone 5 mg/day. The psychotic treatment prescribed was olanzapine 20mg/day, due to previous response to this agent. A small venous angioma confined to right frontal site was detected on MRI and planned to be managed by follow-up visits of 6 month intervals. In contrast to low hemoglobin levels, triglyceride, LDL cholesterol and creatinine levels were found to be high. By the tenth day of hospitalization her voice hallucinations lessened and delusions had stopped. She was free of voice hallucinations by the twenty-fifth day. Her antipsychotic treatment ceased by the fourth month due to lack of psychotic symptoms, weight gain and a positive response to Sheehan syndrome therapy. The patient has been discharged and did not exhibit any psychotic symptom through an 8 month period, but then began to talk about paranoid delusions with her husband. When the patient was asked about medication compliance, she reported taking 50mcg/day less than her prescribed 150 mcg/day dose of levothyroxine. After adjustment for her levothyroxine dose she recovered and her delusions disappeared.

DISCUSSION

In this article, we presented a 44 year old female patient who presented with a psychotic syndrome, and was subsequently diagnosed with Sheehan syndrome 24 years after her initial hemorrhage during child birth, and responded well to treatment.

Hypopituitarism is an endocrine disease characterized by the absence of one or more hypophyseal hormones. In panhypopituitarism, all of the adenohypophyseal hormones are lacking, thus Sheehan syndrome is a form of panhypopituitarism caused by severe hemorrhage during delivery and subsequent hypophysis necrosis (Sheehan 1937).

The symptoms of Sheehan syndrome encompass a broad spectrum since many hormonal failures exist. Agalactorrhea, amenorrhea, intolerance to cold, constipation, drying of skin and weight loss are well known symptoms. This syndrome may also be accompanied by psychotic disorders in some cases. Anorexia nervosa (Danowski et al. 1972), depression (Lynch et al. 1994) and psychotic disorders (Dissanayake and Leiberman 1969) are all implicated among accompanying diseases in hypopituitarism.

There is not a clear relation between hypopituitarism and psychotic disorders yet. Recent studies have particularly focused on an endocrine etiology of schizophrenia. Some studies suggest low levels of estrogen (Riecher-Rössler et al 1994) and thyroid (Othman et al 1994) hormones as an etiology of schizophrenia. Low cortisol levels as seen in Addison disease (Malu et al 1988) or high cortisol levels (Yıldırım et al 2011) are both linked to psychotic disorders. The acute droppings of hormone levels in the postpartum phase are strongly linked to postpartum psychosis. In Sheehan syndrome, levels of cortisol, estrogen and thyroid hormones are decreased and these are implicated among main factors for psychotic disorders.

Psychotic disorders which are related to a general medical event must be timely matched by this event. In our case, it is not definite that the symptoms started with the medical event. Contrary to many Sheehan syndrome linked psychosis syndromes in the literature, there are rare cases of late onset psychotic diseases in Sheehan syndrome (Columbano 2011). Thus it is difficult to establish a sure correlation for our case. However, our patient's story, with a particularly positive response to hormonal therapy, supports presence of Sheehan syndrome as an influence in this patient's psychosis.

Some studies suggested immune impairments for etiology of schizophrenia (Kinney et al. 2010). Respective to grade of tissue necrosis, Sheehan syndrome may cause symptoms within years or days. Also, this tissue necrosis may initiate an autoimmune process which has been reported by some studies (Goswami et al 2002). Sheehan syndrome patients are found to have anti-hypophysis antibodies in 63% of patients, as

compared to 14% in the normal population (14%) (Goswami et al 2002). There are some examples of late onset Sheehan syndrome cases in the literature, one patient had progressive symptoms of amenorrhea and fatigue for 24 years (Nishiyama 1993). In our case, the late presentation of the patient to the clinic scene may be a reason for delayed diagnosis.

The factors including an appropriate response to endocrine therapy, presence of regular perceptions, attending affect and presence of self care are all supporting features of the linkage between psychosis and Sheehan syndrome in our case. Moreover, after adjustment of her levothyroxine dose, she reported remission of her paranoid delusions. With combined treatment of prednisolone and levothyroxine, the patient experienced a subsequent clinical response, thus supporting the presence of a Sheehan syndrome related disease. Also, the role of thyroxine on paranoid disorders may be a focus of new studies.

As a conclusion, autoimmune and endocrine diseases may be concomitant events or contributors to disease in psychotic disorders. The time fashion between medical pathology and psychotic disorders must be considered. Also, the acquisition of a detailed history, physical examination and medical investigations contribute to the proper diagnosis of psychiatric diseases.

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