EUROPEAN JOURNAL OF BIOMEDICAL AND PHARMACEUTICAL SCIENCES
http://www.ejbps.com

ISSN 2349-8870
Volume: 3
Issue: 5
659-661
Year: 2016

SHEEHAN'S SYNDROM AND ACUTE PSYCHOTIC DISORDER: A RARE CASE REPORT

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Article Received on 20/03/2016 **Article Revised on 09/04/2016** **Article Accepted on 30/04/2016**

SUMMARY
Sheehan syndrome is a rare disorder of ischemic pituitary necrosis due to severe post partum hemorrhage. Abnormalities of hypophyseal arteries including external compression, vascular spasm and thrombosis are implicated. Most commonly patient often present with history of post partum lactation failure and amenorrhea with features including tiredness, intolerance to cold, constipation, weight gain, hair loss and slowed mentation, bradycardia and hypotension. Another presenting feature is symptoms of secondary adrenal insufficiency like fatigue, weight loss, hypoglycemia, anemia and hyponatremia. Diagnosis is often delayed. Studies concerning the etiology of psychotic symptoms refer to endocrine and autoimmune systems. In this study, we discussed a case coming with psychotic disorder in which the diagnosis of Sheehan's syndrome was made along with etiological aspect, including the follow-up period and treatment.

INTRODUCTION
Sheehan's syndrome (SS) is a rare disorder of ischemic pituitary necrosis due to severe post partum hemorrhage (PPH). It may present in post-partum period or several years after delivery.[1]

Although it is a well described entity, but unfortunately it is still under diagnosed in developing countries, given that these nations have high proportions of home conducted deliveries. In the pathogenesis of SS, various factors have been proposed like autoimmunity, enlargement of pituitary gland, disseminated intravascular coagulation and small sized sella.[2] Abnormalities of hypophyseal arteries including external compression, vascular spasm and thrombosis have been implicated in SS. In some cases, a woman might be relatively asymptomatic, and the diagnosis is not made until years later when patient present with features of hypopituitarism.[3] Such features include secondary hypothyroidism with tiredness, intolerance to cold, constipation, weight gain, hair loss and slowed mentation, bradycardia and hypotension. Another presenting feature is symptoms of secondary adrenal insufficiency like fatigue, weight loss, hypoglycemia, anemia and hyponatremia. Such a woman may, however can develop adrenal crisis when her body is stressed by various factors like severe infection or surgery years after her delivery.[3]

CASE
Thirty nine year old female patient, housewife by occupation, reported to our department of endocrinology accompanying her family members with recent onset of paucity of speech and movements. She was unable to stand and seemed disoriented to time place and person. There was excessive sleepiness and lack of interest towards family members and personal hygiene. This was associated with repeated episodes of vomiting for last twenty days. She was already being evaluated by a neurologist and was referred for psychiatric evaluation. Patient was married at nineteen years of age and mother of two children. Her first parity at age of 20 years was without any complications. Delivery of second child was full term normal delivery in hospital when she developed severe PPH, severe enough requiring hysterectomy. This was followed by lactation failure. On detail enquiry, her family members revealed that patient had severe weakness and frequent body pains including arthralgias and myalgias for which she got frequent analgesics and calcium supplementations. There was no past history of any psychiatric or neurological illness in her or any of her family members. She was neither a smoker nor a drinker and had never abused illicit drugs. On general examination, she looked pale and anemic; brittle hairs, forehead wrinkling, breast atrophy was present with loss of axillary hairs. She was afebrile and vital signs were normal. She was oriented to time, place and person. Neurological examination was insignificant except unable to walk due to clumsy gait. Mental status examination revealed her appearance as unkempt,

apathetic and uncooperative. Her speech was largely incomprehensive. Her memory was impaired. She had no insight and had no understanding of why she was brought to hospital and her social judgment was poor.

Laboratory investigations including renal function, liver function as well as workup for inflammatory and infectious conditions did not reveal any abnormalities. Her hormonal profile revealed low levels of serum fT4 with inappropriately normalized TSH. S. cortisol was low. S. estradiol was also low with inappropriately normal FSH. Hyponatremia was present. Cholesterol was high with normal triglycerides(Table 1). MRI brain showed empty sella(Figure 1).

Thus the diagnosis of Sheehan's syndrome was confirmed. Patient was put on thyroxine 25 mcg with hydrocortisone 50 mg intravenous stat and 6th hourly and patient showed significant improvement within day 1 with improvement in serum electrolytes and then was later shifted to oral prednisolone 5 mg/day and thyroxine 25 mcg/day. Patient was given counseling about continuation of treatment for her life time and importance of maintaining a regular follow-up. Stress advice was also given to patient.

Table 1: Showing biochemical profile of the patient

	Patients value	Normal range
S. Cortisol	3.34 nmol/l	138-690 nmol/l
Free T4	0.23ng/dl	0.61-1.123 ng/dl
TSH	0.7µIU/l	0.34-5.6 µIU/l
FSH	3.76 µIU/ml	3-13 µIU/ml
S. Estradiol	3.5pg/ml	130-370 pg/ml
S. Sodium	124 meq/l	135-145 meq/l
S.Potassium	4.1meq/l	3.5-5 meq/l
S. Calcium	7.7mg/dl	8.1-10.4 mg/dl
Total cholesterol	256mg/dl	<200 mg/dl

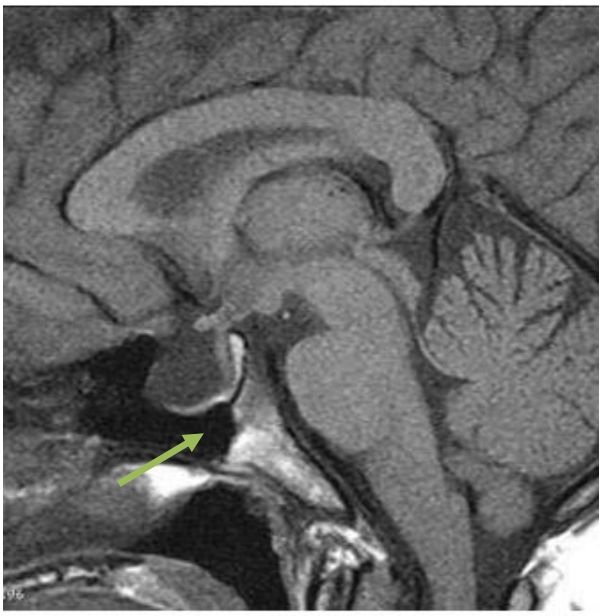

Figure 1: MRI showing empty sella (pointed by arrow)

DISCUSSION

The diagnosis of Sheehan's syndrome is based on history of PPH followed by complete anterior pituitary failure with demonstration of empty sella on MRI pituitary.[4] In this article, we presented a 39 year old female patient who presented with a psychiatric syndrome, and was subsequently diagnosed with Sheehan syndrome 18 years after her initial hemorrhage during child birth. Sheehan's syndrome can present in the immediate postpartum period or after many months to years following delivery. A study of 60 patients has shown the average time between the previous obstetric event and diagnosis of SS to be 13 years.[5] There are some examples of late onset SS cases in the literature; one patient had progressive symptoms of amenorrhea and fatigue for 24 years.[6] In our case, the late presentation of the patient to the clinic scene may be a reason for delayed diagnosis Patients usually present as failure to lactate or amenorrhea following delivery, genital and axillary hair loss, asthenia and weakness, fine wrinkles around the eyes and lips, signs of premature aging, dry skin, hypo pigmentation and other evidence of hypopituitarism which was also present in our case. The absence of amenorrhea or the presence of postpartum lactation, however, does not rule out the diagnosis.

Although patient with SS can present with wide spectrum of psychiatric manifestations ranging from anorexia nervosa[7], depression [8] and psychotic disorders[9], it has received little attention due to rarity of the disorder in the western countries. Various studies are now carried out focusing on the endocrinological etiology of psychiatric complaints. Low estrogen levels[10] and thyroid [11] hormones have been implicated in schizophrenia. Similarly low cortisol levels as seen in Addison disease [12] or high cortisol levels [13] are both linked to psychotic disorders. Moreover acute droppings of hormone levels in the postpartum phase are strongly linked to postpartum psychosis.

Immune impairments in SS have also been suggested for etiology of schizophrenia. [14] Respective to grade of tissue necrosis, Sheehan syndrome may cause symptoms within years or days. This tissue necrosis may start an autoimmune process which has been reported by some studies.[15] Sheehan syndrome patients are found to have anti-hypophysial antibodies in 63% of patients, as compared to 14% in the normal population.[15]

We present a case of acute depression in a patient of SS. She developed Sheehan's syndrome preceded by postpartum hemorrhage. There is a sudden drop in the levels of hormones leading to a relative deficiency of these hormones during postpartum which may be responsible for some of the psychiatric symptoms. [16]

Contrary to various SS linked psychosis syndromes in the literature, there are rare cases of late onset psychotic diseases in Sheehan syndrome. [17] In our case, patient had vague complaints of body aches with low mood

followed by sudden onset of severe depression following vomitings. Dyselectrolytemia and acute adrenal insufficiency might explain etiology of sudden onset acute depression. Also improvement in symptoms following hormonal therapy supports presence of SS as an influence in this patient's psychosis. Treatment with thyroxine and glucocorticoids resulted in complete recovery after attaining euthyroid and eucortisolemic state.

CONCLUSION

Depression and other psychotic disorders are uncommon in Sheehan's syndrom. Autoimmune and endocrine etiologies might be concomitant events or contributors to disease in psychotic disorders. This case report emphasizes the importance of a meticulous history taking & examination and medical investigations in making proper diagnosis of psychiatric diseases.

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