

Case Report

Successful Management of Patient with Sheehan's Syndrome Presenting with Psychosis and Catatonia

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ABSTRACT

Sheehan's syndrome is a neuroendocrine condition that manifests with symptoms of hypopituitarism. It mostly occurs as a complication of parturition due to severe postpartum hemorrhage compromising pituitary circulation. Reports of neuropsychiatric manifestations of Sheehan's syndrome are available with most cases describing psychosis. We report an interesting case of Sheehan's syndrome which presented with catatonia on a psychotic background and its successful management. The possible pathophysiological underpinnings for the causation of catatonia and psychosis in this condition are also discussed.

Key words: *Catatonia, hypopituitarism, psychosis, Sheehan's syndrome*

INTRODUCTION

Sheehan's syndrome is a condition where hypophysial necrosis occurs in a setting of severe hemorrhage and profound hypovolemia. It is most commonly encountered following severe postpartum hemorrhage (PPH) that compromises pituitary circulation leading to pituitary apoplexy. The condition was first described by an English pathologist H. L. Sheehan who found pituitary necrosis in autopsy brain specimen in females who suffered obstetric shock. The prevalence of this condition is around 2.5%–4% in women above 20 years in a study conducted in India.^[1]

The syndrome manifests with clinical features of hypopituitarism such as lactation failure, amenorrhea, genital and axillary hair loss, asthenia, signs of

premature aging, and dry skin. It can also present as an emergency condition with circulatory failure, severe hyponatremia, diabetes insipidus, hypoglycemia, and congestive heart failure.^[2] There are some reports of Sheehan's syndrome presenting as psychosis^[3,4] or depression.^[5]

We report an unusual case of 43-year-old female presented with catatonic and psychotic symptoms who was later found to be suffering from Sheehan's syndrome and difficulties faced in management.


CASE REPORT

A 43-year-old married female attended the Psychiatry Department with complaints of not speaking and

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How to cite this article: Nath S, Ranjan R, Mohapatra D, Mishra BR. Successful management of patient with sheehan's syndrome presenting with psychosis and catatonia. *Indian J Psychol Med* 2018;40:276-9.

Access this article online	
Website: www.ijpm.info	Quick Response Code 
DOI: 10.4103/IJPSYM.IJPSYM_280_17	

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eating anything, decreased emotional reactivity, urinary incontinence, standing or sitting for hours, and maintaining particular limb positions for a longer duration for the past 2 days. According to her husband, she also expressed fearfulness, suspicion, and claimed to hear some voices for the past 8–10 days before the current presentation. She has two children, the last one being born 13 years back out of normal unassisted home delivery during which she had severe PPH that warranted hospitalization. She had a failure of lactation after delivery, and she never conceived thereafter (secondary infertility). Since then (she was 31-year-old then) till now her menstruation stopped for which no medical treatment was sought.

She was admitted for managing her catatonic symptoms which compromised her food and water intake. A quick physical examination revealed her blood pressure to be low (80/60 mmHg) with cool extremities on palpation (axillary temperature 35.7°C) and her skin appeared to be coarse and dry. Her facial features revealed pallor, slight facial puffiness along with mild loss of hairs in the medial aspect of her both eyebrows. She also lacked axillary and pubic hairs, and her breasts were atrophic. Her respiratory and gastrointestinal system examinations were within normal limits while neurological examination revealed rigidity. On mental status examination, she was found to be unkempt with ill-maintained eye contact and having a blunt affect. She showed catatonic signs, namely, mutism, posturing, and negativism (*gegenhalten*). Due to the lack of speech output, a thorough assessment of her mood thought, perceptual functioning, and higher cognitive functions could not be done. Busch Francis Catatonia Rating Scale (BFCRS) score was 14 at the time of admission. Blood investigations revealed microcytic, hypochromic anemia with hyponatremia [Table 1]. Hormonal assessment was planned since we suspected hypopituitarism which revealed low values of all anterior pituitary hormones [Table 1]. Magnetic resonance imaging was done which revealed partially empty sella [Figure 1]. Considering secondary amenorrhea, loss of secondary sexual features, secondary infertility, unstable vital status along with a history of PPH with reduced anterior pituitary hormones on serum assay and MRI finding of partially empty sella, a provisional diagnosis of hypopituitarism due to Sheehan's syndrome with catatonia was made and she was planned for a detailed workup.

She was started on intravenous fluids for her hypotension which brought her vitals to normal but there always remained a propensity for hypotension. She was administered ½ ampoule intravenous lorazepam stat which dramatically improved her catatonic symptoms (BFCRS score = 5). After this, she was kept

Table 1: Blood investigations and hormonal profile

Blood Investigation	Patient Value
Hb	7.2 g/dl
Total leukocyte count	6630/cu.mm
Platelet	2 lakhs 73 thousand/cu.mm
Peripheral blood smear	Anaemia with microcytic hypochromic RBC morphology with slight anisocytosis
Serum ferritin	220 ng/ml
Fasting blood sugar	76 mg/dl
Na ⁺	128 mmol/L
K ⁺	3.6 mmol/L
Urea	31 mg/dl
Creatinine	1.1 mg/dl
Serology (HbsAg, anti-HCV, HIV-I and II, VDRL)	Negative

Hormonal profile		
Hormone	Normal value	Patient value
ft3 (pg/ml)	230-619	1.3
ft4 (ng/ml)	0.7-1.53	0.22
TSH (μIU/ml)	0.34-5.5	1.28
LH (μIU/ml)	15.9-54.0	3.09
FSH (μIU/ml)	23.0-116.3	11.21
Prolactin (ng/ml)	1.8-20.3	1.37
Cortisol (8 am sample) (μm/dl)	4.3-22.5	1.29

RBC – Red blood cell; Hb – Hemoglobin; HbsAg – Hepatitis B surface antigen; HCV – Hepatitis C virus; VDRL – Venereal disease research laboratory; TSH – Thyroid-stimulating hormone; LH – Luteinizing hormone; FSH – Follicle-stimulating hormone; ft3 – Free triiodothyronine; ft4 – Free thyroxine



Figure 1: Magnetic resonance imaging (brain) showing partially empty sella with thinned out anterior pituitary

on oral lorazepam 3–4 mg/day in divided doses along with tablet olanzapine 10 mg gradually decreased to 5 mg considering a psychotic history as mentioned above. Oral hematinics were also started for her anemia, and a general medicine consultation was sought for managing her hypopituitarism.

Under the general medicine care, she was given parenteral hydrocortisone (100 mg 8 hourly) along with oral thyroid hormone replacement (50 μg/day later increased to 75 μg/day) along with oral fluconazole

for some superficial fungal infections. Her catatonic symptoms improved with lorazepam and her psychotic symptoms improved with the above hormone replacement along with olanzapine 5 mg. She was discharged on olanzapine 5 mg/day, levothyroxine tablet 75 mcg/day, fluconazole 150 mg/day, and hematinic tablet. A follow-up consultation is planned after 1 month.

DISCUSSION

Our case is probably one of the few cases reported depicting catatonia in Sheehan's syndrome globally. On literature search, we could find only single case which was also reported from India^[6] the diagnosis of Sheehan's syndrome is made in our case considering a history of PPH followed by lactation failure, secondary amenorrhea and infertility, lack of secondary sexual characteristics (atrophic breasts, loss of sexual hairs) along with a hormonal profile depicting hypopituitarism. Other associated features such as anemia, hyponatremia, hypoglycemia, hypothermia like in our case are also documented in Sheehan's syndrome.^[2] The astonishing factor here is the late diagnosis (13 years) since the vascular event. The catatonic manifestations and current hospitalization along with somatic pointers paved the way for unearthing the diagnosis which would have otherwise been still camouflaged. Previous reports have also pointed to a delay in years in diagnosing this condition.^[7]

Earlier reported cases of psychosis in this condition mentioned management difficulties, but none of them presented with catatonic symptoms. Shoib *et al.* reported psychosis in a lady with Sheehan's syndrome that failed to respond to olanzapine but showed good response to hormonal therapy.^[3] Similarly, Kale *et al.* reported of postpartum psychosis in a case of Sheehan's syndrome which improved with hormone replacement only.^[4] A recent report by Reddy *et al.* showed psychosis marked by both delusion of persecution and auditory hallucination in a 42-year-old female who did not respond adequately to antipsychotic (risperidone, later switched to quetiapine) but showed satisfactory response to hormone replacement in conjunction with the antipsychotic^[8] the patient in our case showed 2 days of catatonic symptoms on a background of psychosis which brought her to clinical attention. The catatonia resolved dramatically with lorazepam thus stressing the fact that the line of catatonia management is not different to catatonia due to other causes. The addition of hormone replacement to olanzapine in our case improved her psychotic symptoms as like the earlier case which still emphasizes the need for hormone replacement therapy in this condition.

No specific pathophysiological link has been reported between Sheehan's syndrome and psychosis and neither for catatonia. A role of hypothyroidism^[9] and hypoestrogenism^[10] has been earlier proposed for schizophrenia. A similar link between hypocortisolemia^[11] and hypercortisolemia^[12] with psychosis has also been proposed. Similarly, sudden lowering of hormones in the postpartum period has been implicated in postpartum psychosis. Considering the above evidence, it appears apparent that the hyposecretion of these hormones due to pituitary necrosis is probably implicated in the causation of psychosis. Their role in catatonia in this condition is still doubtful, and there remains a need to elucidate this link in experimental studies. Considering the role of lorazepam in the treatment of catatonia and consequent causative role of gamma-aminobutyric acid in catatonia, there may be some connection with this inhibitory neurotransmitter between Sheehan's syndrome and catatonia.

Whatever may be the causative link and its treatment implication, catatonia, and psychotic symptoms, in this case, was a boon in disguise since these symptoms paved the way for a proper diagnosis of Sheehan's syndrome. We propose a comprehensive and holistic management in catatonia cases with a thorough physical examination and relevant laboratory investigations so as to guide proper treatment of individual case on its merit.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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