

Cushing's syndrome with Unusual Presentation: A Case Report

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Abstract: Cushing's syndrome may present with a wide spectrum of symptoms but is less recognized that psychiatric symptoms can form part of the presenting clinical features. Cushing's syndrome needs to be considered if there are features like hirsutism, truncal obesity, abnormal glucose tolerance etc. Here we report a case of 32 year old female who presented with delusional ideas along with physical features like obesity, hirsutism. Prompt diagnosis not only crucial for psychiatric management but also important for timely initiation of necessary management for hypercortisolemia. Several investigations were performed like fasting and postprandial blood glucose, Thyroid profile, and Cortisol and ACTH estimation along with Magnetic Resonance Imaging of brain. She was treated with Amisulpride, Metformin, Glimepiride, Ketoconazole, Spironolactone and responded well with the treatment.

Keywords: Cushing's syndrome, Hirsutism, Delusion, Cortisol, ACTH

INTRODUCTION

The relationship between endocrinology and psychiatry has gathered a good deal of attraction among health care professionals. Endocrine disorders may some time demonstrate prominent mental abnormalities. In majority of the cases psychiatric abnormalities follow the manifestations of endocrine disorders. There may be situation where psychiatric manifestation gain prior attention. Among patients reported from general hospital settings, psychiatric manifestations have been found in more than 50% of the cases of Cushing's syndrome [1]. The diagnostic hazard lies with these patients who develop psychiatric features in initial period of illness. These may demonstrate the picture in such a way that endocrine abnormality goes unnoticed.

Endogenous Cushing's syndrome results from chronic excess glucocorticoid production from the adrenal glands. It may be adrenocorticotrophic hormone (ACTH) dependent (80%-85%) or independent (15%-20%). ACTH dependent causes include most commonly a pituitary corticotrophic adenoma, less frequently an extra pituitary tumor and rarely a tumor secreting corticotrophic releasing hormone (CRH)[2]. Hyper vigilance is necessary because patients can present with features that can be non specific and insidious in development. The non psychiatric clinical features of Cushing's syndrome includes central adiposity, proximal myopathy, thin skin, purple striae on the trunk, hirsutism, menstrual irregularity, glucose intolerance etc. Depression, mood dysregulation, sleep disturbance and cognitive abnormalities are observed in Cushing's syndrome [2]. Major depressive syndrome is seen most commonly in (50%-70%) of the cases [1]. Other associated features include anxiety and hypomania [3,4]. Psychosis and mania are the less common features [5,6]. The severe psychosis accompanying Cushing's syndrome are mostly

depressive in nature. Patients may also develop a disorder in which persistent or recurrent delusions dominate the clinical picture. Delusions may be accompanied by hallucinations. Features suggestive of Schizophrenia such as bizarre delusions, thought broadcasting may also present in some cases.

Evaluation of the patients with suspected Cushing's syndrome is complex. Initial screening test includes 24 hour free urinary cortisol level and or lack of cortical suppression after low dose dexamethasone testing. Once hypercortisolemia is confirmed ACTH level, CRH stimulation test, Magnetic resonance imaging of brain should be done.

Here we report a case of 32 year old female who presented with delusional ideas along with physical features like obesity, hirsutism, menstrual irregularity, impaired glucose tolerance. Several investigations were performed like fasting and postprandial blood glucose, Thyroid profile, Cortisol and ACTH estimation along with Magnetic Resonance

Imaging of brain. All these were done for an appropriate and early diagnosis which will help in the proper management and to understand the pathophysiology of this rare presentation.

CASE REPORT

A thirty two year old Hindu unmarried female from urban area attended the Psychiatry out patient department with her parents. Her parents complained of her persistent belief that she was being married and having a family with her husband. Her parents also reported that when they tried to convince, she became annoyed and did not accept the truth that she was not married. She repeatedly said that she got married with a person who was her neighbour at some point of time after a long term relationship. She also reported several events about their married life which was unreal. Her parents informed that all these mental symptoms started from last three and half years back and initially she complained of headache and low mood. She became irritable and aggressive from time to time when her parents opposed her view. During the interview process her parents reported that she was having low mood for some period of time when she was searching for her husband. They also reported that she had developed a tendency to wear colorful sarees and tried to groom herself as a married lady. Her family members informed that she had gained her body weight along with menstrual irregularity for for last four and half year. She developed increased facial hair that is hirsutism from last four years along with other virilising features.

On examination she was obese having increased facial hair growth with mild palor and she was normotensive.(Fig:1,2) During mental status examination she was oriented to time place and person and was in well kempt state but rapport was difficult to establish in the initial part of interview. She was having irritable to low mood from time to time and was having a delusional idea about her marriage with a particular person. There was no history of any perceptual abnormality and judgmental capacity was impaired and she was having grade I insight with complete denial of illness.

Detail investigation was done and hematological report showed Hemoglobin of 9.8 gram% with normal thyroid profile. Her fasting blood sugar was 160 mg/dl and postprandial blood sugar was 220 mg/dl. Hormonal investigation report showed raised cortisol level in low dose dexamethasone suppression test along with high twenty four hour urinary cortisol level. Her ACTH level was 61.75pg/ml (Ref range 9-52 pg/ml), which was raised. Magnetic resonance imaging of brain do not reveal any abnormality. Most of the ACTH secreting tumor are less than 5 mm in diameter and half them are undetectable by imaging. She was diagnosed as Organic delusional disorder as there was clear temporal relationship between the development of underlying

disease that was ACTH dependent Cushing's disease and the onset of mental symptoms.

She was treated with antipsychotic Amisulpride starting with a low dosage from 50 mg and increased up to 400 mg. Amisulpride is better antipsychotic to prevent weight gain, metabolic syndrome. She was treated with Ketoconazole, Spironolactone for the management of ACTH dependent Cushing's syndrome after consultation with Endocrinology department. She was advised to take Metformin and Glimepiride to control blood sugar abnormalities. She responded well with the treatment and psycho-education was given to her family members and was advised to continue medication and regular follow up.



Fig-1: Hirsutism and Typical Face



Fig-2: Facial Hair Growth (Hirsutism)

DISCUSSION

In this particular case we had found delusional ideas in a patient with florid features of Cushing's disease and all these mental symptoms started after the development of Cushing's disease. Conceptual models have been reported to try to explain glucocorticoid actions in the brain. Psychiatric features of Cushing's syndrome are associated with increased net glucocorticoid signaling via the glucocorticoid receptors. Changes in cortisol activity leading to decreased serotonin and increased dopamine cerebral activity have also been described [7]. The physical and biochemical features associated with Cushing's syndrome can be varied and subtle at times. Psychiatric symptoms may be the original presentation and the

diagnosis can easily be missed without a high index of suspicion. Psychiatric symptoms can potentially be refractory to psychotropic medications with untreated hypercortisolemia, and the underlying cause can have a significantly high mortality and morbidity if left unrecognised.

It is evident that early recognition and timely institution of appropriate management can minimise morbidity and mortality significantly in such type of rare presentation.

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