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•CASE REPORT•

"Neuro-psycho- BS": A Case Report of Rare Association with Bipolar Disorder

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Summary: Bechet's Disease is an inflammatory disease characterized by recurrent oral ulcers (OU), genital ulcers (GU) and uveitis which can develop a neurobehavioral syndrome, also defined as 'neuro-psycho- BS'. Depending on vascular or parenchymal lesions, the presentations could be varied. Due to sparse literature, there is no consensus on management of psychiatric illness comorbid with Bechet's disease. Bipolar disorder in Bechet's disease is extremely rare. Here, we are presenting a case of episodic exaggeration of Bipolar disorder along with Bechet's disease which imposed both clinical and management challenges.

Key words: Bechet's Disease; Neurobehavioral; Bipolar Disorder.

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1. Introduction

Bechet's syndrome (BS) is a systemic, chronic, relapsing vasculitis, typically characterized by recurrent orogenital ulcers, ocular inflammation and skin manifestations; articular, vascular, gastroenteric and neurological involvement may also occur.[1] Besides the other clinical features of BS, it seems relatively frequent that patients with BS develop a neurobehavioral syndrome, characterized by euphoria, bipolar disorders and paranoid attitudes, loss of insight/disinhibition, and indifference to their disease, defined as 'neuro-psycho-BS'.[2]

Bechet's disease and bipolar disorder share features like relapsing and remitting course and viral triggers have been reported for both.[3] In autoimmune diseases affective symptoms have been noted and autoantibodies are reported to accompany affective syndromes.[4] An autoimmune pathogenesis as one of the possible pathogenesis was proposed for unipolar or bipolar disorders.[3] Another possible mechanism could be due to NBD parenchymal damage in the brain.

2. Case report

A 49 year old gentleman developed Bechet's syndrome at the age of 38 years.The illness was episodic in nature with intermittent remission and exacerbations. The symptoms were characterized initially by slurring of speech, gait disturbance, visual disturbance, right side drooping of the eyelid, swelling and redness of periorbital region, recurrent aphthous lesion in the mouth along with weakness of the lower limbs and unsteady gait. He was found to be HLA-B51 leucocyte antigen positive.

For the past 4 years along with the above said symptoms which were also episodic in nature, the patient presented with episodic illness with symptoms characterized by euphoric mood, over familiarity,

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increased goal-oriented behavior, increased self-esteem, grandiose delusions, overspending, increased libido and exhibited disinvited behavior. Currently he presented with 2 months of relapse of similar symptoms such as euphoric mood, reduced need for sleep, over activity, increased libido, over spending, increased energy level and over religiosity affecting bio-socio-occupational function followed by exaggeration of Bechet's syndrome even with good medical compliance. Patient was not on any mania inducing drug to suspect iatrogenic causes. Basic laboratory profile is included in table 1. Likewise, he had three similar relapses and all were followed by exaggeration of Bechet's disease. His Young mania rating scale (YMRS) point was recorded as 35 at the time of admission. The patient was started on valproate 1000mg/d and quetiapine 400mg/d. The patient showed improvement in both his manic and psychotic symptoms in the next 3-4 weeks. YMRS was again assessed at this point of time and was recorded as 4. The patient was also initiated on methylprednisolone 50mg/day which was gradually tapered and stopped over the next 7 weeks and azathioprine 75 mg/day which the patient continued to take post-discharge as maintenance treatment for the primary illness which was necessary to prevent both medical as well as psychiatric relapses.

3. Discussion

Behcet's disease usually starts around the third or fourth decade of life. The characteristic inflammatory lesion in BD is secondary to involvement of large and small arteries as well as venous vessels. Around 5-20% of the population with BD also exhibit neurological complications, and are defined as neuro-Bechet's disease (NBD).[5]

Neuro-Bechet's disease underlying pathology is divided into vascular and parenchymal lesions. Patients with vascular lesions display intracranial hypertension with papilledema, headaches, focal neurological deficits and occasional pyramidal symptoms. In contrast, parenchymal lesions cause pyramidal symptoms in 96% of patients and behavioral alterations in half. A minority also suffers from hypersomnia, sensory and gait aberrations and hearing and sight problems. Only 2% of cases exhibit psychiatric symptoms, such as acute psychosis.[6]

Bipolar disorders have been described in rare cases. In 2002, Aydin et al. reported a case of neuro-BS in which the initial onset was hypomania.[7] To date, there have been no guidelines for treatment of these disorders in BS, probably because bipolar disorders have been studied less frequently than other types of disorder in BS patients. Indeed, these symptoms may be related to a neurological substrate of the syndrome, and we can hypothesize that they develop in the middle stage of the disease. A combination of mood-stabilizing drugs, such as sodium valproate, carbamazepine and olanzapine, may produce some improvement in the disease.[1]

In our patient there were both neurological and psychiatric symptoms with parenchymal involvement that matches with the imaging findings and mood episodes exaggeration of Bechet's symptoms. It's unclear whether the psychiatric symptoms are due to the underlying NBD or independent, as evidence

Table 1. Assessment and results

ASSESMENT	RESULT
CNS EXAMINATION	B/L PERIORBITAL SWELLING. LEFT EYE-PTOSIS, IMPAIRED TAMDEM GAIT
MMSE	30/30
NEURO-PSYCHOLOGICAL ASSESMENT(NIMHANS Neuropsychology Battery-2004)	INVOLVEMENT OF DLPFC AND RIGHT TEMPORAL LOBE
YMRS	35 at admission and 4 after four weeks of admission
HLA-B51 LEUCOCYTE ANTIGEN	POSITIVE
BLOOD INVESTIGATIONS	NEGATIVE FOR ANA PROFILE, RA FACTOR, VDRL, SERUM NMO,HBS,HCV
MRI BRAIN	Multiple focal lesions in basal ganglia, internal capsule, thalamus and deep white matter of frontal lobe. Round lesion in right temporal lesion Multiple well defined demyelinating lesions.

MMSE: Mini-Mental State Examination; YMRS: Young Mania Rating Scale

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from literature on the causal relationship between psychiatric symptoms and NBD is limited.

Only a few case reports have linked NBD with bipolar disorder, one reported manifestation of mood disorder during an NBD exacerbation.[8] Deniz O et al. reported a psychotic patient with an NBD exacerbation and a patient whose affective symptoms preceded BD.[9] Due to the lack of literature we might assume that the psychiatric disorder in our patient could be caused by NBD parenchymal damages in the brain. Also, there is an association between bipolar disorder and inflammatory cytokines. Studies have proved that increased CRP level may predict manic episode in a depressive patient. Also, there were multiple active inflammatory markers in men with bipolar illness.[10] Organ specific autoimmunity like thyro-peroxidase antibodies (TPO-Abs), antibodies to H/KAT-Pase , GAD65A, antibodies to gliadin have been linked with Bipolar disorder as well as unaffected relatives of bipolar disorders.[11] Then there is also a lack of literature about the causal relationship between the two and little research has been done so far with respect to treatment of NBD with mood disorders.

Combination of glucocorticoids and azathioprine along with mood stabilizers and anti-psychotics appears to be effective as it showed good results in our patients although specific guidelines are not available.

Further research is required to understand the casual relationships, and is needed from a treatment prospective as well. This case report is just an eye opener in this sparsely researched area.

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All authors contributed to the drafting of this manuscript

"神经精神综合征"：一例与双相情感障碍相关的病例报告

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概述：白塞氏病是一种炎症性疾病，特点是复发性口腔溃疡（OU）、生殖器溃疡（GU）、和葡萄膜炎，该疾病可以发展为神经行为综合征，也定义为"神经精神综合征"。根据血管或器质性病变，其表现可以是多样的。由于文献稀缺，有关共病白塞氏病的精神疾病的管理还没有达成共识。白塞氏病中的双相情感障碍是非常罕见的。在这里，我们陈述一个有关共病白塞氏病的双相情感障碍加重的案例，并且实施了临床和管理方面的挑战。

关键词：白塞氏病；神经行为；双相情感障碍

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