

Kliver-Bucy Syndrome in a Patient with Bipolar Affective Disorder: A Case Report

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ABSTRACT:

Introduction: Kliver and Bucy described a behavioral syndrome in rhesus monkeys following bilateral temporal lobectomy which included psychic blindness, hyperorality, hypermetamorphosis, hypersexuality, and emotional unresponsiveness. **Case report:** A 44 years old right handed male of Indo-aryan origin, blacksmith by profession, had presented in manic phase of bipolar illness. He had hypersexuality, hypermetamorphosis, hyperorality, and altered dietary habits along with amnesia and fleeting misrecognition of even his close relatives. MRI of the patient showed mild cerebral atrophy with right temporal lobe atrophy. The patient was treated with lithium and olanzapine along with benzodiazepines. The symptoms resolved gradually with resolution of the manic phase. Patient had similar features in the previous manic episode as well that resolved with resolution of mania. **Conclusion:** The symptoms of Kliver-Bucy syndrome like increased libido and increased activity might be confused with that of mania. Other features of Kliver-Bucy syndrome and the overt hypersexuality could help identify it even during manic phase of bipolar illness.

Keywords: bipolar affective disorder • BPAD • Kliver-Bucy syndrome • mania

INTRODUCTION:

Kliver and Bucy (1939) described a behavioral syndrome in rhesus monkeys following bilateral temporal lobectomy which included psychic blindness, hyperorality, hypermetamorphosis,

hypersexuality, and emotional unresponsiveness. [1] Complete or partial presence of symptoms of the syndrome have been reported on and off in human since then. We present a case of Bipolar Affective Disorder (BPAD) with manic episode presenting with features of Kliver-Bucy Syndrome (KBS) as the patient had recurring features of KBS during manic phase, resolving with resolution of the manic episodes. So, we are reporting the case to inform the clinicians of such unique findings of transient and recurring KBS in case with BPAD.

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CASE REPORT:

Case presentation:

A 44 years old right-handed male, educated up to class three, blacksmith by occupation, from low-socioeconomic Hindu family was admitted with ten years of BPAD, episodic with full inter-episodic remission, with past two episodes of mania and one episode of depression. The current episode was fourth one with one and half month duration, acute onset, and progressive deterioration after three



months of non-compliance with lithium 900 mg/day.

Patient presented with predominantly irritable mood with increased talkativeness, grandiose talks, increased energy level and activity, physically assaultive (provoked and unprovoked), social disinhibition, decreased need of sleep, and difficult to control at home. He gradually started expressing overt sexual desire, exposing his genitalia and physical attempts at coitus even to minor girls and close relatives. He raped a six year old girl and even killed a small goat of bleeding due to coitus. He used to try to have coitus with animals and inanimate objects as well. Patient had to be restrained multiple times because of his disinhibited behavior and violence but when let free, would immediately start exhibiting hypersexuality. He used to touch everything that comes on his way, pulling things and snatching them. He would keep both edible and inedible things into his mouth, even his own feces when physically restrained. He had fluctuating impaired recognition of even his close relatives. He also had impairment of recent memory.

He did not have any other chronic medical problem and no history of similar or other major medical illness in the family.

Patient had history of one manic episode and one depressive episode in the past, details of which could not be obtained. He had history of one similar episode two years back and was admitted in the same hospital and managed with lithium carbonate and olanzapine. However his features of KBS were missed as they were less severe and confused as symptoms of mania itself.

Clinical Findings:

Patient was admitted and managed in the psychiatry ward of university teaching hospital. His physical examination findings were within normal limits. Patient had hypersexuality expressing overt sexual gestures and hip shaking mimicking copulation even during physical restraining. He had sniffing at almost everything that he gets hold of and putting them into mouth and discarding them immediately suggestive of hyperorality. He was found even putting discarded vaginal pads into mouth. He would also try to touch anything around him and get hold of it suggestive of hypermetamorphosis. Patient was predominantly irritable with frequent rage episodes without any apparent provocation and required frequent chemical and physical restraint, both for his rage and hypersexuality. He had delusion

of grandiose identity and power, and had increased energy level. He also had decreased need for sleep. Patient had impairment of recent memory but intact immediate and remote memory. His social, personal, and test judgment were impaired and he denied having any illness.

Diagnostic Assessment:

On examination, his general physical condition was within normal limits. On mental status examination, patient had increased psychomotor activity, hypermetamorphosis, hyperorality, and hypersexuality. Speech was increased in tone, volume, and production with pressure of speech at times. Predominant mood was irritable. Patient had grandiose delusion. No apparent hallucinatory behavior was noted. He was at times disoriented to time and place but oriented to person. Recall after three minutes and recent memory was impaired. He had concrete thinking on proverb testing and impaired social test and personal judgment. He did not have insight about his illness. His investigations of Complete Blood Count, Random Blood Sugar, Urea, Creatinine, Electrolytes, Liver Function Tests, Thyroid Function Tests, and EEG were within normal limit. Serology for HIV, HCV, and HBV were non-reactive. MRI brain showed mild diffuse brain atrophy with prominent CSF spaces in the medial part of right temporal lobe likely to be due to atrophy (Fig 1 and Fig 2). Bender-Gestalt Test (BGT) done after admission had score of 10, strongly suggesting organicity.

Patient's overt hypersexuality, high level of hyperactivity, and putting almost everything into mouth was not being explained by manic phase alone. So, we reviewed the history again that clearly established hypersexuality, hyperorality, hypermetamorphosis, rage episodes, fleeting misrecognition of family members, and similar history in the past manic episode.

The findings of BGT strongly suggested organicity and the findings of right temporal lobe atrophy in brain MRI further helped us to make the syndromal diagnosis of KBS. We could not however identify any etiology of the patient's right temporal lobe atrophy. There was no history of any seizure, head trauma, and tuberculosis, and no features of any other neurodegenerative or metabolic disorders could be established. So, the final diagnosis of Bipolar Affective Disorder Manic Episode with Psychotic Symptoms with Kluver-Bucy Syndrome



Fig 1: T1W axial section showing prominent temporal horn of right lateral ventricle along with prominent CSF spaces in the ipsilateral medial temporal lobe (hippocampal/parahippocampal region) suggestive of atrophy of medial temporal lobe. Arrow shows the dilated and prominent CSF space in the right medial temporal lobe

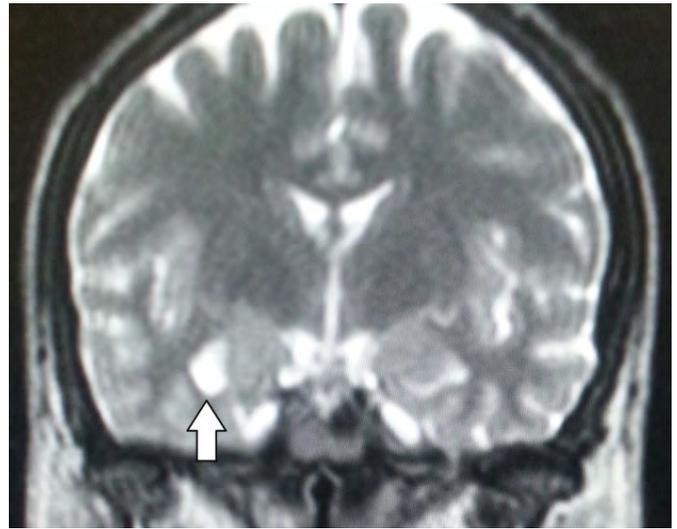


Fig 2: T2W coronal image of brain with arrow showing asymmetric dilated CSF space in the right medial temporal lobe

was made. Timeline of the patient's history, work-up, and interventions is presented in Table 1.

Therapeutic Intervention:

Patient was started on Lithium 900 mg/day, olanzapine 10 mg/day gradually optimized to 25 mg/day and lorazepam six mg/day. Patient also

Table 1: Timeline of the patient's history, diagnostic work-up, and interventions

Dates	History	Diagnostic Testing*	Interventions
Ten years back	History suggestive of depressive episode	none available	not known to informants and no available documents
Five years back	History suggestive of manic episode requiring hospitalization but details not known as patient was working abroad.	none available	not known to informants and no available documents
Two Years back	History of manic episode with features of KBS. Admitted in the same hospital for one month and discharged. Diagnosis of KBS missed that time as the symptoms were confused with that of mania.	CBC, electrolytes, TFT, LFT, urea, creatinine, blood sugar, chest x-ray were within normal limits. Serology for HIV/HCV/ HBV non reactive.	Managed with lithium carbonate 900 mg/day and olanzapine 30 mg/day. Olanzapine was gradually tapered off over one year and continued on Lithium 900 mg/day
Three months back	Patient became non-compliant citing no illness.		
On presentation	Manic symptoms along with hypermetamorphosis, hyperorality, hypersexuality, transient amnesia, fleeting misrecognition of family members and rage episodes. Patient improved gradually with resolution of KBS symptoms in two weeks and manic symptoms in three weeks. He had hyperphagia for the initial weeks gaining seven kg in 11 days that subsided gradually. At the time of discharge in three weeks patient had complete amnesia of his behavior during the acute phase.	CBC, electrolytes, TFT, LFT, urea, creatinine, blood sugar, chest x-ray were within normal limits. Serology for HIV/HCV/ HBV non reactive. EEG done was within normal limits. MRI brain showing mild cerebral atrophy with right temporal lobe atrophy.	Managed with lithium 900 mg/day and olanzapine 25 mg/day. Patient required injection haloperidol 10 mg with injection promethazine 50 mg deep IM TID along with physical restraint on and off for initial one week.
Follow up upto two years	Patient's olanzapine was gradually tapered off over one year but he had persistent hypersomnolence for one and half year after which it subsided on its own without any further intervention. Patient was fully functional after that.	Follow up MRI scan of brain in two years showed no significant further changes in the brain.	Lithium 900 mg/day continued.

* CBC - complete blood count; TFT - thyroid function test; LFT - liver function test; EEG - electroencephalogram

required parenteral haloperidol upto 30 mg/day for initial one week along with physical restraints for his hypersexuality and rage episodes. His olanzapine was gradually tapered off over one year and Lithium 900 mg/day was kept as the mainstay of treatment. Patient was also suggested various behavioral interventions for his hypersomnolence, but without much benefit.

Follow up and Outcomes:

Patient's symptoms of KBS gradually improved over two weeks duration and manic symptoms resolved by almost 90% in three weeks. Patient had anorexia at the time of admission but developed hyperphagia after admission, gaining seven kilogram (57 to 64 Kg) weight in 11 days which gradually subsided with improvement of his illness. On improvement, patient had amnesia of his previous behavior and it persisted even upto two years follow up. Immediate and remote memory was intact.

Patient was followed every three months and he was compliant to medicines. His follow up MRI brain in two years revealed the same findings as the previous one and there were no major changes. His Thyroid Function Test (TFT) results done every six months were normal until two years follow up. Patient became fully functional after one and half years but had persistent amnesia of the acute phase even after two years.

DISCUSSION:

The case initially appeared to be an usual case of BPAD in manic phase. But his overt hypersexuality and hyperorality prompted us to review the diagnosis and we could establish KBS in this case. We could also establish similar features of KBS in one of the previous manic episodes, which was missed in our setting itself. So, any atypical features presenting in any psychiatric cases should prompt us to look for probable organicity and alternative diagnoses. We could not get more detailed history of the initial two mood episodes. We also could not establish the etiology of the right temporal lobe atrophy.

The classic presentation of Kluver-Bucy syndrome was produced experimentally in monkeys in lab by bilateral temporal lobectomy.[1] However, in human, such discrete bilateral temporal lobe lesions cannot be reproduced. So, the presence of the classic features of KBS as originally reported is rarely observed. So, diagnosis of KBS in humans

could be made if any three of the following symptoms are present - hyperorality, hypersexuality, hypermetamorphosis, visual agnosia, changes in emotional behavior, and changes in dietary habits. [2] The change in emotional behavior as originally reported was placidity of affect and lack of fear response. However, Lilly et al. have reported of aggression and rage in one of their cases.[2] Various etiologic causes have been described in the literature ranging from herpes encephalitis,[3] to post-traumatic dementia,[4,5] tubercular meningitis,[6] SLE,[7] epilepsy,[8] intracranial hemorrhage,[9] and others.

We reported this case here because some of the features of KBS could be confused with that of manic episode. In our case, we could pick up KBS when the increased libido in the patient was so much excessive unlike that of usual cases of mania. Increased activity of manic episode could also be confused with that of hypermetamorphosis of KBS; however, our case not only had distractibility and increased activity but also had marked tendency to touch, hold, pull, or push things around unlike that of distractibility of mania. KBS in human have also been said to be almost invariably associated with aphasia, amnesia or dementia.[2] Our case had amnesia during the acute phase and amnesia of the acute phase was persistent even at two year follow up. The other differential diagnoses to be considered in this case are paraphilias, temporal lobe epilepsy, and Obsessive Compulsive Disease (OCD) with sexual contents. In our opinion, had the sexual abnormalities occurred in isolation, paraphilia could have been the provisional diagnosis. However, our case had mainly severe hypersexuality that included both animate and inanimate targets - from children to adults in human, any animals (goat, cows) and inanimate objects. Besides, paraphilic disorders are usually insidious in onset, chronic and enduring in nature while our case had acute onset, short lasting and recurring pattern. Similarly, mood disorders and OCD can be co-morbid conditions and OCD can be accentuated during acute phase of BPAD. But, OCD is also a chronic illness and persists even after the resolution of acute phase of BPAD. In our case, there were no features suggestive of OCD prior to or after the resolution of acute phase making it unlikely to be part of OCD. Sometimes, temporal lobe epilepsy could have such presentation. However, in our case, the behavioral problems had lasted for almost seventy days, had been mostly persistent throughout

and the episode resolved without use of antiepileptic drugs. Keeping in view the protracted course, the behavioral manifestations could not be explained either by episodes of temporal lobe seizure or by post-ictal behavioral problems alone.

Whatever the etiology, almost all cases of KBS have involvement of the temporal lobe. Our case had mild diffuse cerebral atrophy with atrophy of the right medial temporal lobe on MRI of the brain. Any atypical features in any psychiatric patients should prompt us to look for organicity and alternate diagnosis.

Patient perspective:

The patient had no memory of the acute phase and was reluctant to acknowledge his behavior during the acute phase initially. However, later, he acknowledged that he might have done those things though he does not remember and was worried if it would recur. He was thus compliant to medications and other behavioral interventions.

CONCLUSION:

In our case, the symptoms of KBS has been partial and transient, recurring during manic episode of bipolar illness. It is important to note that hypersexuality and hypermetamorphosis in KBS might be confused with increased libido and increased activity of manic episode. Thus, other associated features of KBS should be looked for.

Competing interests:

The author declare that no competing interests exist.

Financial disclosure:

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