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Case Report

Rare presentation of Klüver-Bucy syndrome following subarachnoid hemorrhage

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ABSTRACT

Background: Klüver-Bucy syndrome (KBS) is a rare neuropsychiatric disorder, and it can be associated with a variety of neurological disorders. It is characterized by visual agnosia, placidity, hyperorality, hypersexuality, dietary changes, amnesia, and hypermetamorphosis. KBS is mainly a clinical diagnosis, with at least three symptoms sufficient to diagnose the condition.

Case Description: The case describes a 49-year-old Filipino woman with a history of hypertension who presented with symptoms strongly suggesting KBS following subarachnoid hemorrhage, including behaviors such as hyperorality, hypermobility, placidity, hypermetamorphosis, and hypersexuality along with memory disturbance. She was managed as a case of brief psychotic disorder initially with olanzapine, then on the second presentation as a case of delirium with risperidone.

Conclusion: Among many symptoms of KBS, only three symptoms are required for the diagnosis clinically. Numerous neurological conditions can cause KBS. Symptomatic treatment is the mainstream treatment currently for KBS.[3] While different differential diagnoses are present, neurologists, psychiatrists, neurosurgeons, and radiologists should collaborate and be vigilant for the diagnosis of KBS, especially with the presence of one of its etiologies.

Keywords: Delirium, Hyperdocility, Hypersexuality, Klüver-Bucy, Placidity, Psychosis, Subarachnoid hemorrhage

INTRODUCTION

Klüver-Bucy syndrome (KBS) is a rare neurobehavioral disorder associated with the disturbances of temporal portions of limbic networks that interface with multiple cortical and subcortical circuits to modulate emotional behavior and affect.[4] The full syndrome is rarely seen in humans because the anterior temporal lobe dysfunction is usually less severe in humans when compared to that following total temporal lobe resection in monkeys. Hypersexuality was the most common symptom, followed by hyperorality.[2] Other symptoms are placidity, visual agnosia, hypermetamorphosis, memory deficit, prosopagnosia, and hyperdocility.[2] We are presenting a case of a 49-year-old female patient who developed KBS secondary to subarachnoid hemorrhage.

CASE PRESENTATION

A 49-year-old female, known to have hypertension but with no history of mental illness, works as a housemaid and is functioning well. She was referred from the primary healthcare center to the ER at King Fahad University Hospital, Saudi Arabia, on April 14, 2020, after she was observed for 1 day by her sponsor talking to her dead husband and taking her passport outside the house, asking to go back to her country. In addition to her auditory hallucinations, her sponsor reported that she tried to seduce him, which suggested hypersexuality. During the interview, she was disoriented to place. She also exhibited delusional thoughts, believing that her dead husband was Canadian and still alive. She was admitted to our psychiatric ward for observation, and computed tomography (CT) scan showed calcification in the hippocampus [Figure 1]. She was diagnosed with a brief psychotic disorder and was started on 5 mg olanzapine and discharged on April 19, 2020. The patient stated that she continued taking olanzapine for 2 weeks afterward.

On December 26, 2020, the patient presented again to the hospital complaining of headaches for 3 days and one seizure attack. On examination, the Glasgow Coma Scale fluctuated from 13–14/15 [Figure 2], the patient was sleepy and ambulating with assistance. The investigations report stated CT scan showed subarachnoid hemorrhage with ventricular enlargement, and computed tomographic angiography showed a left posterior communicating artery aneurysm [Figure 3]. Cerebrospinal fluid (CSF) analysis reported a pinkish/red color, red blood cell count of 1777 without any white blood cells, protein 22.8, glucose 137, and negative CSF Gram stain. The external ventricular drain was inserted, followed by coiling of the aneurysm [Figure 4], and then nimodipine 60 mg was started. Repeated CT further showed interval mild progression in size in the lateral and third ventricles. The patient underwent permanent shunt due to hydrocephalus secondary to subarachnoid hemorrhage [Figures 5 and 6].

On December 31, 2020, psychiatry consultation was done. The patient complained of sleeplessness and continuous auditory hallucinations. She denied any depressives, anxiety, or delusional symptoms. It was on and off, disoriented to time, place, and person. The provisional diagnosis was delirium, and she was prescribed 1 mg risperidone at bedtime. Throughout her stay in the hospital, she exhibited hyperorality by persistently insisting on eating even when she had nothing by mouth (NPO) status. The nurses reported that the patient started to show childlike behavior and seemed "as innocent and kind as a child," which seemed like placidity. In addition, the sponsor noted her increased kindness, expressing a desire to care for and serve others, which suggested hyperdocility. Furthermore, she was confessing about things she did and asking for forgiveness.

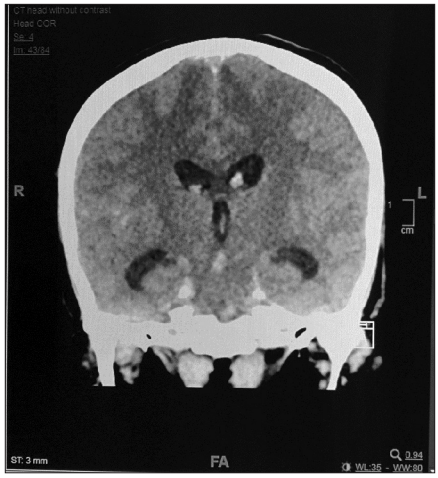

Figure 1: Hippocampus calcification.

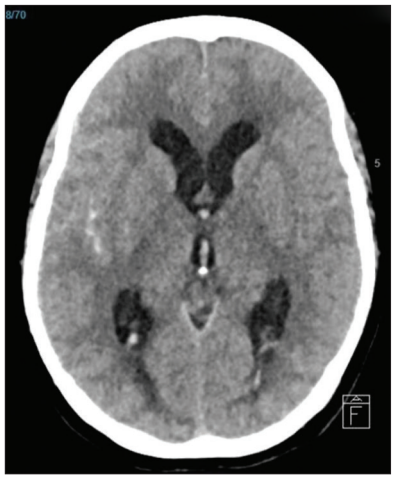

Figure 2: Initial computed tomography showing subarachnoid hemorrhage.

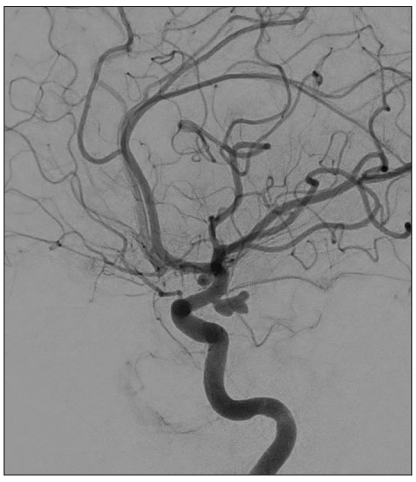

Figure 3: Aneurysm pre-coiling.

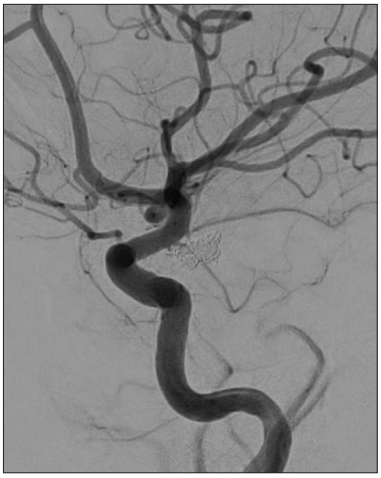

Figure 4: Aneurysm post-coiling.

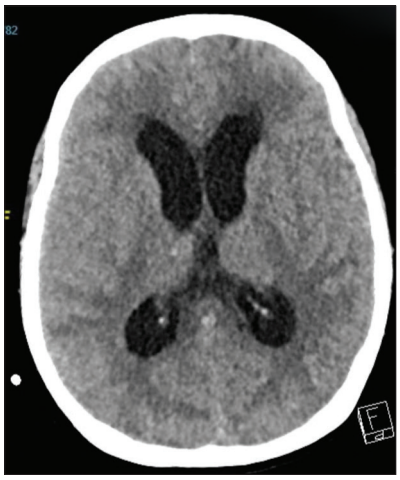

Figure 5: Computed tomography showing hydrocephalus.

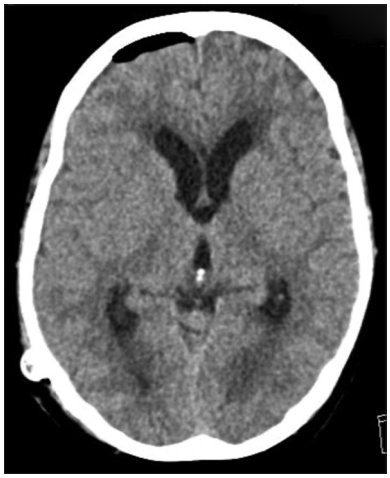

Figure 6: Computed tomography after shunt placement.

Moreover, the patient approached to grasp and touch the female examiner's hand to feel its temperature and asked the examiner twice during the same interview about how the examiner was doing, despite the examiner replying to the patient the 1st time, which could suggest hypermetamorphosis and memory disturbance, respectively.

Consequently, the patient's symptoms were strongly suggestive of KBS resulting from subarachnoid hemorrhage. On January 19, the patient was discharged on 1 mg risperidone. We lost follow-up with her as she traveled back to her country.

DISCUSSION

The exact prevalence of KBS is difficult to estimate; case series and reports are <200 reported cases in the literature.[1,3] It is a clinically diagnosed syndrome that can be easily missed or misdiagnosed, whereas at least three symptoms are reported to be sufficient to diagnose the condition. Symptoms are hypersexuality, hyperorality, placidity, visual agnosia, hypermetamorphosis, bulimia, amnesia, prosopagnosia, and hyperdocility.[2] In this case, we observed many symptoms of KBS: hypersexuality, hyperdocility, hyperorality, placidity, memory disturbance, and possibly hypermetamorphosis. Some of the symptoms, for example, hypersexuality, appeared before other symptoms with a period of seeming stability in between; it seems that it could be due to antipsychotic use, specifically olanzapine 5 mg daily. However, we do not have enough input to contradict or confirm this information as the compliance is questionable. While there could be an associated pathology such as delirium, we still cannot rule out mental disorders such as brief psychotic disorder or manic episodes, noting that there was no family history of mental disorders. There are many different causes of KBS. The most common ones are head trauma, bilateral temporal infarction, and herpes simplex encephalitis in children. The less common pathologies are subarachnoid hemorrhage, encephalopathy, encephalitis, Alzheimer's disease, Pick's disease, porphyria, hypoglycemia, and adrenoleukodystrophy.[5] In the first presentation of the patient, her CT scan showed calcification in the hippocampus along with subarachnoid hemorrhage, which is one of the most common radiological findings in KBS.[3] There is only symptomatic treatment for such cases with KBS.[3] In this case, we used risperidone to manage the symptoms. The most common medications used in the management of KBS secondary to traumatic brain injury (TBI) are mood stabilizers, antidepressants, and antipsychotic drugs. Carbamazepine and leuprolide, in particular, are used to decrease hypersexuality, while haloperidol and anticholinergics are effective in managing the behavioral abnormalities associated with KBS. In 50% of KBS following TBI cases, the patients fully recovered from the disorder.[2] One-half of all these cases had pharmacological management, with carbamazepine being the

most commonly prescribed medication that showed promising results.[2] As with subarachnoid hemorrhage, patients can present with behavioral changes that should alert the physician to the specific KBS combination and to be vigilant to it. Therefore, it is undeniable that early detection and proper management of KBS, whether secondary to epileptic seizures, infections, post-infectious causes, TBIs, or subarachnoid hemorrhage, can lead to a better prognosis.[3]

CONCLUSION

As KBS is a rare neuropsychiatric disorder. Collaboration between the treating neurologist, psychiatrist, neurosurgeon, and radiologist is crucial for establishing the final diagnosis of KBS. The definitive treatment of KBS remains a challenge due to the complex presentation of the disorder, and the definitive recommendation to guide clinical practice is still in need of further research.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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