We present a case with frontal lobe symptoms and Klüver-Bucy-like syndrome following subarachnoid hemorrhage and hydrocephaly. Klüver-Bucy syndrome is a rare neurobehavioral condition characterized by placidity, visual agnosia, hypersexuality, hyperorality, and hypermetamorphosis (the tendency to react to or to touch every visual stimulus). The syndrome is usually associated with lesions of the amygdala or its pathways, and it occurs after head trauma, anoxia-ischemic encephalopathy, herpes simplex encephalitis, and Reye’s syndrome. A 45-year-old right-handed female patient, who developed hydrocephaly after meningitis due to bilateral middle cerebral artery aneurysm surgery presented to our psychiatry clinic with various behavioral and emotional changes. In her psychiatric examination, increased and disinhibited speech, perseveration, placidity, impaired go/no go task performance, and hyperphagia were observed. The patient was treated with risperidone 0.5 mg/day. Magnetic resonance imaging (MRI) of the brain showed encephalomalacic-gliotic changes in the anterior superior medial temporal lobe (including bilateral amygdala), hydrocephalus, bilateral abnormal signal intensity in the white matter of the frontal region, and bilateral infarction in the centrum semiovale.

Symptoms, such as placidity (loss of anger and fear) and altered dietary habits are some of the clinical features of Klüver-Bucy syndrome, whereas disinhibition and perseveration are associated with prefrontal cortex dysfunction.

**Key Words:** Klüver-Bucy Syndrome, frontal lobe, amygdala, magnetic resonance imaging

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**INTRODUCTION**

The relationship between temporal and frontal lobe injury, and psychiatric signs and syndromes has been known for many years. Klüver and Bucy (1938, 1939) described behavioral changes, which could be generated in monkeys, through bilateral temporal lobectomy (amygdala, hippocampus, and adjacent cortical structures). These animals had the tendency to inspect all objects with their mouths instead of their hands. This is known as psychic blindness or visual agnosia. These animals also had decreased motor or vocal reactions to fear- and anger-provoking stimuli, increased interest in any object entering their visual field (hypermetamorphosis), a tendency to eat edible and non-edible things in large quantities, and increased sexual activity (hypersexuality). Three of the above-mentioned symptoms are required for the diagnosis of Klüver-Bucy syndrome (Lilly et al., 1983). Even though there is a general impression that favors the view that amygdala damage must be bilateral and that the syndrome is not observed with unilateral damage (Lilly et al., 1983), in single case reports it has been shown that similar symptoms can be observed after left temporalectomy (Ghika-Schmid et al., 1956) and right amygdala damage (Yoneoka et al., 2004).

Diseases, such as herpes simplex encephalitis, anoxic-ischemic encephalopathy, cranial trauma, Pick’s disease, transtentorial herniation, adrenoleukodystrophy, Reye’s syndrome, CO poisoning, and subdural hemorrhage, have been included among the causes of Klüver-Bucy syndrome in humans (Lilly et al., 1983). It was previously reported that Klüver-Bucy syndrome was not specific to amygdala damage and that similar signs could be observed in frontal lobe injury (Takahashi and Kawamura, 2001) or isolated diencephalon lesions (Müller et
al., 1999), a phenomenon which has been thought to be caused by damage to the connection pathways between the dorsomedial thalamus, prefrontal cortex (PFC), and other limbic structures.

Full-blown Klüver-Bucy syndrome is rarely observed in humans, and similarly, the clinical presentation of the symptoms can vary. Symptoms such as difficulty differentiating facial expressions (prosopagnosia), placidity or blunted affect, hypermetamorphosis, hyperorality or excessive eating, hypersexuality or changed sexual behavior have been described (Lilly et al., 1983).

Frontal lobe injury is a more common clinical entity. Cognitive processes, such as planning, execution, step-wise thinking, sequencing, judgment, strategy shifting, elasticity of behavior, volition, insight, visualization, foresight, and working memory, are frontal lobe functions (Mesulam, 2000). There are numerous interconnections between striatum, the limbic system, and primary-secondary cortical areas. The emergence of symptoms in the areas of cognition, perception, and emotion is related to disruption of the frontal-striatal-thalamic circuit. Damage to this circuit forms the basis for disorders such as obsessive-compulsive disorder and frontal personality disorder (Joseph, 1999).

In the presented case, brain imaging data and behavioral changes observed after a subarachnoid hemorrhage and its complications will be discussed.

**Disease history**

A 45 year-old right-handed female patient, with no known history of systemic disease, was brought to the hospital after fainting at home and subsequently underwent surgery because of a subarachnoid hemorrhage (SAH) caused by bilateral middle cerebral artery (MCA) aneurism bleeding. During the first postoperative week the patient had high fever. Lumber puncture (LP) was performed and the patient was diagnosed with meningitis, which was treated with antibiotic therapy. A control cranial computed tomography (CT) at the time revealed enlarged ventricles. LP was repeated and the opening and closing cerebrospinal fluid (CSF) pressures were measured 195 mmH₂O and 145 mmH₂O, respectively. At the end of the fourth week of hospitalization, the patient was discharged with phenytoin and metoprolol treatment and a follow-up plan for hydrocephalus. Based on the husband’s report, the patient had undulating partial loss of consciousness episodes, which lasted almost 10 days. Partial loss of consciousness was accompanied by ataxia, and urinary and fecal incontinence. At the end of this 10-day period, symptoms, such as partial loss of consciousness and incontinence, had improved.

Subsequently, meaningless talking was observed, which lasted almost one month. At the time, CSF pressures, as measured during the control LPs, were normal. The patient was claiming that she had 3 grandchildren, even though there was only one grandchild at home, and she was treating her daughter-in-law meanly, even though they had not previously had any problems. For 3 days the patient said that she had seen her house being stoned, and that she was afraid of being harmed. The patient was generally tense and anxious. She was not calmed down by the reassurances of her relatives and she repeated her fear of evil things that could happen to her. At home, she was continuously talking to herself. She was referred to the psychiatry outpatient clinic by the department of neurology due to these symptoms. During the first psychiatric interview the patient was continuously talking to herself, her place-person-time orientation was intact, and she had delusions of persecution and visual hallucinations. Risperidone 2mg/day was started based on the diagnosis of psychotic disorder. During the follow-up period her fears, anxiety, and delusions of persecution subsided, and meaningless talk decreased. Her relations with her family members also improved. However, the patient was observed to be talking incessantly and unnecessarily, and continued to repeat the same subjects. Premorbidly, the patient was described as a person who did not talk much, easily got angry, and was very critical with her affairs; however, after the onset of the disease, she became careless with her chores, and her cooking deteriorated. She was no longer setting boundaries during her interactions with other people and she was not behaving according to social norms. She began to intervene inappropriately in on-going conversations and she was asking disturbing questions to people. Her eating habits had also changed. She was inclined to eat all the time without feeling hungry. Even though she was premorbidly described as some one who easily got angry, after the event she had become placid. Her anger at things had subdued. The dose of risperidone was gradually reduced to 1mg/day and then to 0.5mg/day.

Family history: Her mother and one sibling had a history of hypertension and heart disease. There was no family history of psychiatric disorder.

**Examination results**

Systemic examination was normal. Neurological examination revealed the patient had anisocoria. She had
minimal bilateral cog wheel rigidity. The mental status examination found that patient's personal grooming was mediocre. Her speech was comprehensible, but circumferential. She displayed disinhibition. Her affect was euphoric. Perseveration was noted. Thought content was poor, though there were no obsessions or delusions. In terms of cognitive abilities, her attention was normal. Other than for the events at the time of the disease period, her short-term and long-term memory were intact. She was able to remember 2 out of 3 words after 5 minutes. Her orientation to person, time, and place was intact. There were no perception pathologies. Her judgment and reality testing abilities were intact. Calculation ability was normal. She was able to perform simple calculations. Abstract thinking was intact. The level of intelligence was observed to be normal. Frontal functions, especially her performance on the go/no go test, which is a test that specifically evaluates disinhibition, were damaged significantly. The frontal tests were not administered because of the patient’s education level.

**Laboratory examination**

Routine laboratory tests were normal. Results from the brain magnetic resonance imaging (MRI) (Figures I and II) were as follows:

- Encephalomalacic-gliotic changes, including cystic areas in the anterior superior regions of the temporal lobes, bilaterally; artifact due to bilateral MCA clipping.

- Opercular insular tissue damage at the right temporal region.

- Bilateral frontal signal change.

- Multiple periventricular ischemic/gliotic lesions.

- Triventricular hydrocephalus, mild cerebellar atrophy.

- Bilateral infarct regions at the location of the centrum semiovale.

**Diagnostic discussion**

The psychiatric symptoms of the patient were analyzed during 2 different periods in time. The first time window was the postoperative period that lasted almost one month, in which the following symptoms were observed: incoherent talking; uneasiness; irritability; delusions of persecution; visual hallucinations; relationship difficulties. During the second time window the psychotic symptoms had improved, specific changes in personality had emerged, and the following symptoms were observed: disinhibition; perseveration; decreased social insight; difficulty in planning; placidity; excessive eating. In this paper the focus will be on the symptoms observed during the second time window.

When the changes in personality were analyzed, it could be seen that frontal and temporal regions had been affected. Symptoms such as perseveration, disinhibition, difficulty in planning, decreased social insight, and inappropriate behavior are markers of frontal lobe dysfunction. The frontal lobe is thought to be the center of executive functions. The limbic system has a critical role in the processing, integration, inhibition, and recall of stimuli coming from the striatum and cortical sensory areas (Joseph 1999). Different clinical presentations have been described in frontal lobe syndrome, based on the anatomic location of the damage: disorganized type (dorsal convexity); disinhibited type (orbitofrontal); apathetic type (mesial frontal) (Duffy and Campbell, 2001). In our case, the symptoms were probably related to an orbitofrontal system lesion. MRI (Figure II) revealed periventricular signal changes in the frontal lobes, bilaterally. It has been reported that the prefrontal cortex (PFC) is the first anatomic area to be affected in hydrocephalus and that the most common symptom is perseveration (Cummings 1995). The lack of atrophy in the frontal and temporal lobes differentiates it from frontotemporal dementia, which has a similar onset to frontal lobe symptoms (Neary et al., 1998). The presented case can also be differentiated from Lewy body dementia, a disorder that presents with progressive loss of visuospatial abilities and frontal lobe functions, in which visual hallucinations and parkinsonism symptoms are prominent, by the results of neuroimag-
ing and by the lack of parkinsonism. Rigidity, which is a parkinsonism symptom, is among the basic symptoms of Lewy body dementia; however, a resting tremor is not frequently observed (Geser et al., 2005). The mild signs of parkinsonism observed in our case emerged after antipsychotic medication use and disappeared after down titration of the medication. In other words, the signs of parkinsonism did not accompany the clinical picture from the beginning.

Our case’s excessive talking, disinhibition, and euphoria were suggestive of a manic episode; however, the lack of other manic symptoms, such as increased associations, flight of ideas, reduced need for sleep, excessive spending, increased libido, and feeling energetic, led us away from such a diagnosis. Another important issue that needs to be emphasized is the observation of changes in affect and eating habits, which seem to be related to the bilateral temporal lobe damage. As can be seen with MRI (Figure I), the sequela that formed as a result of the surgery consists of a bilateral temporal lobe lesion in which the amygdala is included. Premorbidity, the patient had been a person of irritable nature; however, after the incident she became more placid and she began to eat more. These symptoms are among the characteristics of Klüver-Bucy syndrome, which can be seen after amygdala injury.

It is well known that the medial temporal lobe plays an important role in survival, and in such behaviors as eating and reproduction (Mesulam, 1999).

It has been shown that bilateral amygdalectomy performed in mice causes a decrease in freezing behavior and autonomic responses when confronted by a cat. In 2-month-old monkeys, isolated amygdala lesions caused a decrease in anxiety when confronted with lifeless objects; whereas, it caused an increase in anxiety during social interactions (Prather et al., 2001). Pradhan et al. (1998) emphasized the differences between the clinical pictures observed in children (3-6 years) and have emphasized the fact that social learning influences the presentation of symptoms. One study demonstrated that the most frequent behavior changes observed in cases of isolated amygdala lesions are placidity (decreased fear and anger response) and hyperorality (Davis, 1999).

Even though there was a lack of past hypersexuality, because the classical Klüver-Bucy diagnostic triad of hyperorality, hypersexuality, and placidity was not met, it can be said that in the presented case, there were changes of affect and eating habits caused by a bilateral amygdala lesion. The patient’s hippocampus had been relatively spared and there were no significant memory disturbances.

It is rather difficult to speculate retrogradely about the differential diagnosis of the case during the first time window, in which psychotic symptoms had dominated the clinical picture. The fact that the patient could not remember anything about this time window made it difficult to reach a decision. However, based on the outpatient records and the reports of the husband, it can be said that the patient’s intact orientation and lack of symptom undulation throughout the day excluded a diagnosis of delirium. The most probable diagnosis could have been psychotic disorder due to a general medical condition. It seems as if this clinical picture had temporally been related to the hydrocephalus observed after the SAH. Postoperative meningitis and the adhesions caused by it could have facilitated the formation of hydrocephalus.

In conclusion, a case that presented with both frontal lobe and Klüver-Bucy syndrome symptoms, which were caused by injury inflicted upon the frontal and temporal areas due to infection and surgical intervention, has been discussed. Frontal lobe syndrome is a relatively more common condition, whereas its causation by bilateral amygdala lesions is a rarity. This phenomenon is interesting because of its potential to elucidate the functions of the amygdala.
REFERENCES


