Behcet's Disease Presented with Isolated Peduncular Hallucinosis: A case report

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Citation

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Abstract

Bilateral paramedian thalamic infarctions are associated with several neuro-psychiatric manifestations besides from neurological, neuro-ophtalmological and pathological signs. Peduncular hallucinosis is rarely seen among these neuro-psychiatric symptoms. Peduncular hallucinosis is especially associated with upper brain stem lesions in the literature. We report a case of 20- year-old man who had vivid, active, and colorful hallucinations, somnolance, and amnesia. We detected bilateral paramedian infarction in cranial magnetic resonance imaging due to Behcet's disease and discussed the symptoms seen in the lesions of this region.

INTRODUCTION

Thalamic infarction may cause several neuro-psychiatric conditions such as somnolance, behavioral, memory, and orientation disturbances besides neurological and neuro-ophtalmological signs ([1,2,3]). Neuro-psychiatric signs such as loss or impairment of consciousness with sudden onset, apathy, confabulation, amnesia, frontal lobe syndrome, attention deficits and loss of motivation are frequently seen in paramedian thalamic infarcts. These clinical signs are seen more often in patients with bilateral paramedian infarct than those with unilateral infarcts. In the literature peduncular hallucinosis is seen in upper brain stem lesions. It has also been reported in unilateral and bilateral thalamic infarcts ([3,4]). Although changes in mental-psychological status in neuro-Behcet's disease patients were observed and reported, there is no case presented with peduncular hallucinosis associated with Behcet's disease in the literature.

We report a case of bilateral paramedian thalamic infarct due to Behcet's disease presenting psychotic symptoms.

CASE REPORT

A 20-year-old-man was admitted to the emergency department with hallucinations, incoherence, and somnolance. It was learnt from his family that he was in depression, and he felt bad, wanted to sleep and he slept for thirty hours two days ago. He saw dwarf people and strange creatures, and talked inconsistently, and walked continuously in the examination room. His past medical and familial history were unremarkable. He was conscious but disorientated to time in his first examination. He talked quietly with short, meaningless and incoherent sentences. Although immediate and far memory examinations were normal, short and close memory examinations were impaired. He had echolalia and echopraxia. He was smiling with derisive face expression continuously. He said that he had seen his girl friend and naked women images on the wall and started to talk to them during this interview. While the interview was going on, he stood up suddenly and started to look at the walls and made signs with his fingers in the air. When we asked what happened, he stated that there were colorful birds flying and he was shooting at them and said that they did not die but the bullets made holes in their bodies and he did not understand this. The patient was hospitalized in the psychiatry clinic with the pre-diagnosis of psychotic episode because of hallucinations and loosening of association. He had somnolance and disorientation on the other day but his neurological examination was normal. Slight hypodense suspicious lesion was seen in left thalamus on cranial computerized tomography and then magnetic resonance imaging was performed. In this examination hypointense and hyperintense lesions were seen on proton and T_2 weighted sections respectively (Figure 1a and 1b). Therefore he was transferred to the neurology clinic and examined for the etiology of cerebrovascular diseases. The detailed laboratory examination; complete blood count, biochemical tests, electrocardiography, echocardiography,

and MR angiography showed no pathology. The patient was given antiaggregant therapy. He had no change in his memory impairment. He was followed up to six years and nearly total improvement was detected in hallucination, association, somnolance and other cognitive functions. One year after the admission, the patient presented oral ulcers, painful vesicular lesions on lips and oral cavity mucous membrane and glans penis. We detected pustular lesion on both face and extremities. Pathergy test and HLA B 5 tests were positive. The patient was diagnosed as central nervous system involvement of Behcet's disease.

Figure 1

Figure 1: MRI of patient (axial proton weighted echo image) reveals bilateral paramedian thalamic infarct

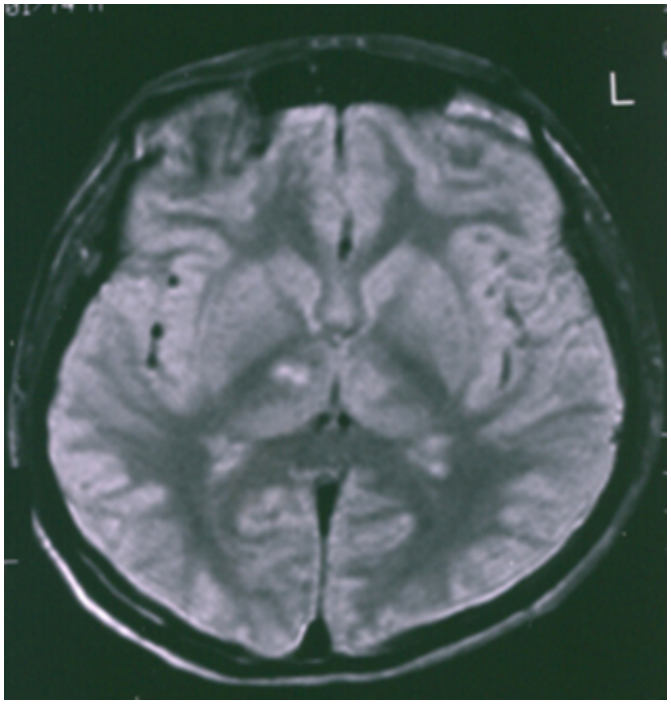
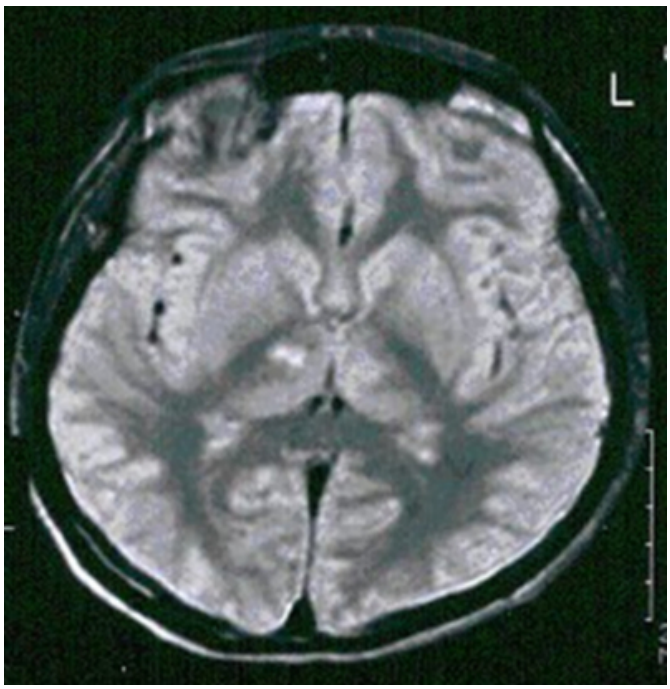


Figure 2

Figure 2: T- weighted axial MRI showed bilateral paramedian thalamic infarcts.



DISCUSSION

Rostral basilar artery occlusions cause neuro-psychiatric findings besides ophtalmological, motor, and sensorial symptoms due to ischemia or infarct in midbrain, thalamus, hypothalamus, paramedian diencephalone, medial temporal lobes. These neuro-anatomic regions are involved especially in neuro-Behcet's syndrome ([5,6,7]). The most interesting one among these neuro-psychiatric disorders is peduncular hallucinosis described in 1924 by Van Bogaert ([1]). These hallucinations were described as active colorful objects having good shapes. The insight was maintained ([1,4]). When the patient with peduncular hallucinosis was examined with autopsy or neuroradiological methods, the lesion responsible for the symptom stated that it had been found in upper brain stem periaqueductal gray matter. The relationship between the lesion and symptom could be explained with release phenomena of reticular activated system ([1,8,9]). However, Feinberg reported peduncular hallucinosis in a case with only right paramedian thalamic infarct without mesencephalic lesion ([4]). Hallucinations in our case were active, vivid, and in natural color and this was convenient with peduncular hallucinosis described in the literature. It was thought that the exaggerated reactions of the patients during their hallucinations could be explained with thalamo-frontal connections ([10]). In our case, no lesion of upper brain stem was observed on cranial tomography and MRI.

Posterior thalamo-subthalamic artery feeds the upper mesencephalon besides the medio-dorsal part of the thalamus ([11]). Somnolance of the patient for a long time, confusion, and memorial impairments at the onset of the disease are classical findings seen in the occlusion of the same artery ([2]) but absence of neurological and ophtalmological findings show that there was no involvement of upper brain stem. This condition made us to think the thalamic structures could be responsible for the pathogenesis of the peduncular hallucinosis. Bilateral involvement of mamillo-thalamic tract in the feeding area of this artery also explains the memorial impairments in our patient ([1,11,12]). When the bilateral thalamic infarcts go with only hallucination and memorial disturbances without motor and ophtalmological symptoms it can be difficult to diagnose.

Neuro-psychological worsening in neuro-Behcet's syndrome can be seen regardless of attacks. Visually and verbally recall memory deficits, disinhibition, apathy, personality changes, executive dysfunction of frontal lobes are neuro-psychological pathologies associated with neuro-Behcet's syndrome ([6,13,14]). These symptoms and signs in cases of Behcet's disease are due to mesodiencephalic involvement.

The fact that a case of Behcet's disease presented with hallucination was found interesting.

References

1. Caplan LR. Top of the basilar syndrome. Neurology 1980: 30 : 72-9
2. Bogousslavsky J, Regli F, Assal G. Thalamic infarcts: clinical syndromes, etiology and prognosis. Neurology 1988: 38:837-48
3. Fukutake T, Akada K, Ito S, Okuda T, Ueki Y. Severe personality changes after left paramedian thalamic infarct. Eur Neurol 2002:47(3):156-60
4. Feinberg WM, Rapcsak SZ. "Peduncular hallucinosis" following paramedian thalamic infarction. Neurology 1989:39:1531 - 36
5. Kocer N, Islak C, Siva A, Saip S, Akman C, Kantarci O, Hamuryudan V. CNS involvement in neuro-Behcet syndrome: an MR study. AJNR 1999:20(6):1015-24
6. Akman- Demir G, Serdaroglu P, Tasci B, and the Neuro-Behcet Study Group. Clinical patterns of neurological involvement in Behcet's disease: evaluation of 200 patients. Brain 1999:122:2171-82
7. Siva A, Kantarci OH, Saip S, Altintas A, Hamuryudan V, Islak C, Kocer N, Yazici H. Behcet's disease: diagnostic and prognostic aspects of neurological involvement. J Neurol 2001: 248:95-103
8. McKee AC, Levine D, Kowall NW, Resell H, Richardson EP. Peduncular hallucinosis due to bilateral infarction of medial substania nigra pars reticulata: A serial-section, postmortem analysis. Ann Neurol 1988:124:125-26
9. Geller TJ, Bellur SN. Peduncular hallucinosis: magnetic resonance imaging confirmation of mesencephalic infarction during life. Ann Neurol 1987:21:602-4
10. Eslinger PJ, Warner GC, Grattan LM, Easton JD. Frontal lobe utilization behavior associated with paramedian thalamic infarction. Neurology 1991 Mar:41 (3):450-2
11. Bogousslavsky J, Regli F, Assal G. The syndrome of unilateral tubero-thalamic artery territory infarction. Stroke 1986:17:434-41
12. Clarke S, Assal G, Bogousslavsky J, Regli F et al. Pure amnesia after unilateral left polar thalamic infarct: topographic and sequential neuro-psychologic and metabolic (PET) correlations. J Neurol Neurosurg and Psychiatr 1994:57:27-34
13. Tanor OO, Kurt BB, Gurvit IH, Akman-Demir G, Serdaroglu P. Neuropsychological follow-up of 12 patients with neuro-Behcet disease. J Neurol 1999: 24:113-9
14. Serdaroglu P, Yazici H, Ozdemir C, Yurdakul S, Bahar S, Atkin E. Neurologic involvement in Behcet's syndrome: a prospective study. Arch Neurol 1989:46:265-9

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