

Ipsilateral hemichorea after striatal hemorrhage

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Abstract

Hemichorea after a striatal lesion of the ipsilateral side of the cerebral hemisphere is a very rare condition. A 72-year-old female presented with right-sided bradykinesia and left-sided hemichorea after a left striatal hemorrhage. In a shape analysis of the brain conducted via MRI, her damaged left basal ganglia was severely contracted whereas her right dorsolateral putamen surface was deformed and hypertrophied. Excessive compensation of the contralateral hemisphere may be a possible mechanism of ipsilateral hemichorea in this case.

Keywords: Ipsilateral hemichorea; striatal hemorrhage; shape analysis

INTRODUCTION

Hemichorea is a hyperkinetic movement disorder that involves one side of the body. Hemichorea is generally associated with a lesion in the contralateral side of the basal ganglia (BG).¹ Hemichorea with a lesion in the ipsilateral hemisphere is a rare movement disorder with an unclear pathomechanism.²⁻⁴ In this report, we describe a case of ipsilateral hemichorea after intracerebral hemorrhage in the left striatum. We performed a statistical neuroimaging analysis of the patient and proposed a possible mechanism for the ipsilateral hemichorea.

CASE REPORT

A 72-year-old female visited the neurology department after experiencing involuntary movements of her left hand. She had suffered from a striatal hemorrhage in the left hemisphere five years previously. She reported that the clumsiness of her right arm was a sequela of the hemorrhage whereas the involuntary movements of her face and left arm had insidiously appeared few years ago after the left striatal hemorrhage. She could not remember the exact onset of the involuntary movements. She was also on medications for hypertension and dyslipidemia. None of her medications were drugs associated with movement disorders such as levosulpiride or antipsychotics. She denied having symptoms suggesting underlying *Lewy* body pathology, such as constipation, REM sleep behavior disorder, or

hyposmia. There were no clinical signs leading us to suspect Parkinson-plus syndromes, such as gaze limitation, severe balance problem, and autonomic dysfunction.

The patient has normal intelligence (minimal status examination score: 28/30). The neurologic examination showed continual irregular dance-like movements of the jaw, tongue, and left hand. The left hand also displayed dystonic posturing in the resting state. Mild rigidity and bradykinesia were noted on the right upper and lower extremities. An examination of the left side of the body reported normal muscle tone and motor speed findings (Video 1).

The brain MRI showed encephalomalacic changes in the left putamen and a microbleed in the left thalamus. No lesions were noted in the left subthalamic nucleus (STN). The right hemisphere showed mild leukoaraiosis without significant ischemic or hemorrhagic vascular lesions (Figure 1A).

To quantitatively assess the morphology of her brain, the brain MRI results were statistically compared with those of 20 age-matched healthy control women (mean age = 70.5 years, standard deviation = 4.07) using two neuroimaging analysis methods (voxel-based morphometry (VBM) and a vertex analysis). The controls were recruited from the MRI database of the author's institution. The institutional review board (IRB) approved the retrospective use of the MRI data (IRB approval number: GCH2018003).

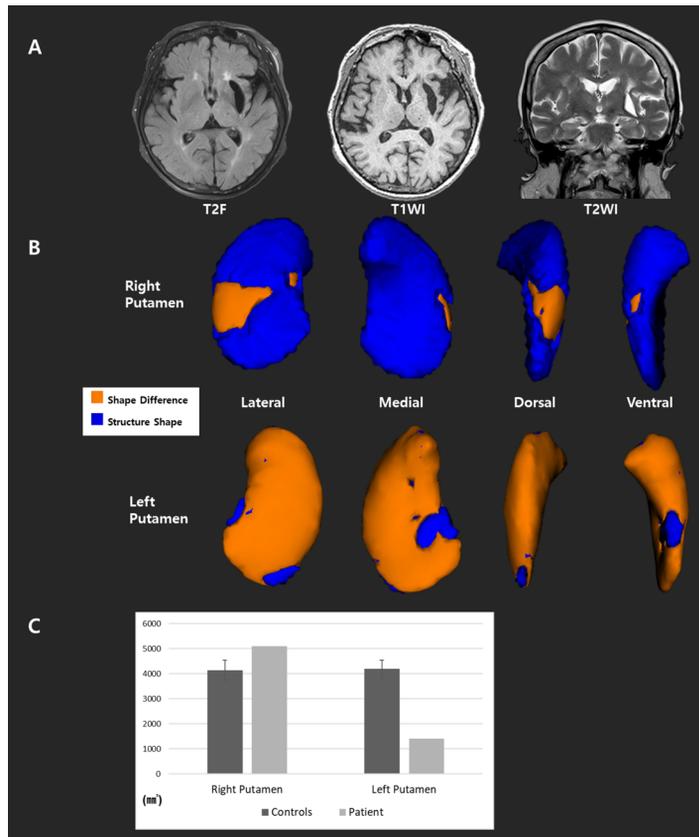


Figure 1. (A) Brain MRI shows encephalomalacic changes of the left putamen. The left subthalamic nucleus is not affected by the lesion (T2F: T2 FLAIR image, T1WI: T1 weighted image, and T2WI: T2 weighted image). (B) The comparison results (controls versus the patient) of the vertex analysis are displayed (orange: significant shape deformation of the patient's brain). A significant shape deformation in the right dorsolateral putamen is identified. Only a small area of the ventromedial surface is saved in the left putamen. A p value of <0.05 was considered statistically significant (corrected for multiple comparisons [family wise error, FWE] using the cluster-wise approach). (C) When the total volumes of the bilateral putamen were quantitatively measured, the volume of her right putamen was greater than one standard deviation above the mean volume of the controls, whereas the volume of her left putamen was severely contracted.

VBM was performed using SPM12 (Wellcome Department of Imaging Neuroscience, London, UK, www.fil.ion.ucl.ac.uk/spm/), which is implemented in MATLAB R2011a (Mathworks, Inc., Sherborn, MA). Gray matter was segmented from the T1-weighted images of the recruited individuals. The segmented gray matter images were normalized into a standardized space. Thereafter, two-sample t -tests were performed to compare the controls with the patient. The shapes of the subcortical structures of the patient's brain were further assessed using FMRIB's Integrated Registration and Segmentation Tool (FIRST), which is part of the FMRIB Software Library (FSL 5.0.8, <http://www.fmrib.ox.ac.uk/fsl>). The subcortical structures, including the putamen, caudate, thalamus, globus pallidus (GP), and

brainstem, were segmented on the brain MRI scans of the patient and controls. A vertex analysis using the FIRST tool was performed to assess the differences in the shapes of the segmented subcortical gray matter structures between the controls and the patient.

VBM did not identify any significant volumetric changes in the cortical areas of the patient's brain. The vertex analysis showed that only a small area of the ventromedial surface of the damaged left putamen was saved. Moreover, the left ventral and dorsal surface of the caudate and the ventromedial GP were partially saved. The dorsolateral surface of the contralateral putamen was deformed and hypertrophied (Figure 1B). There were no significantly deformed areas in the bilateral thalamus, brainstem, right caudate

nucleus, and GP. The measured volume of the patient's right putamen was more than one standard deviation greater than the mean volume of the controls whereas the left putamen was severely contracted (Figure 1C).

DISCUSSION

In this case, the patient presented with right-sided hypokinetic and left-sided hyperkinetic movement disorders after a left-sided striatal hemorrhage. Based on the pyramidal decussation of motor fibers, her left-sided hyperkinetic movements reflected ipsilateral disorder. Ipsilateral hemichorea/ballism is a very rare disorder: only a few cases have been reported in the literature.²⁻⁴ A few explanations on the mechanism behind ipsilateral hemichorea/ballism have been previously proposed, including 1) the release of a previously existing contralateral BG lesion by the ipsilateral hemispheric lesion³, 2) compression of the contralateral hemisphere's vascular system by mass effect⁴, and 3) malfunction of the contralateral STN because of a disruption of the interconnection between the bilateral STN.² In this case, there was no significant deformation of the ipsilateral thalamus, no lesion on the ipsilateral STN, and no definite vascular lesion of the contralateral BG. Therefore, the previous hypothesis regarding the ipsilateral hemichorea was not applicable.

In this report, we performed a statistical analysis on the patient's brain. As expected, her damaged left BG (including the putamen, caudate nucleus, and GP) was severely deformed and contracted. However, the deformed and hypertrophied right dorsolateral putamen was an unexpected finding. Because the dorsolateral putamen is part of the motor loop of the BG, the dyskinesia present in the left hemibody was well correlated with the deformation of the right putamen. The deformation of the right putamen may be a compensatory change in response to the damaged left BG.⁵ Therefore, we can propose another possible mechanism of ipsilateral hemichorea. Because the two BG systems are functionally interconnected, destroying one BG system could place excessive cortical input on the contralateral BG as a compensatory mechanism. The excessive cortical input may have precipitated the ipsilateral hemichorea and caused deformation of the contralateral BG.

However, the current study is limited because it only assessed a single case. Future studies must be conducted to determine the clinical significance

of shape changes in the contralateral BG after unilateral damage to the BG.

DISCLOSURE

Conflict of Interest: None

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Video 1.

[http://neurology-asia.org/content/25/1/neuroasia-2020-25\(1\)-71-v1.wmv](http://neurology-asia.org/content/25/1/neuroasia-2020-25(1)-71-v1.wmv)

In the resting state, her left hand showed dystonic posturing and subtle jerky movements. Shaking of the right hand indicated bradykinesia with mild motor weakness whereas the speed of the left hand shaking was normal. Dyskinesia of her jaw and tongue were also noted during examination. The amplitude of her right arm swing was decreased whereas her left hand showed continuous irregular movements.