Transient Automatic Writing Behavior following a Left Inferior Capsular Genu Infarction

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Key Words

Capsular genu infarction · Automatic writing behavior · Increased writing activity · Perseveration · Abulia

Abstract

A 79-year-old, right-handed woman was admitted to the hospital with decreased spontaneity. Brain magnetic resonance imaging showed a left inferior capsular genu infarction. ^{99m}Tc-ECD single-photon emission computed tomography revealed a left-dominant diffuse hypoperfusion in the basal ganglia and frontal lobe. The patient showed abulia and increased writing activity without motor or sensory deficit. The writing was mainly perseverative, and words written along lines were legible and without spatial distortions. This augmented writing behavior disappeared on day 21. The writing characteristic was more similar to automatic writing behavior than hypergraphia. Dissociation between speech and writing behavior was present in our patient. We suggest that a disconnection within the frontal-subcortical circuit contributed to the development of motor perseveration in writing.

Introduction

Increased writing activity is seldom seen in neurological conditions, except in some patients with cerebrovascular disease [1], frontal-type dementia [2], temporal lobe epilepsy [3], and Parkinson's disease [4]. It has never been reported in capsular genu infarction. We report a patient with a left inferior capsular genu infarction who developed abulia and increased writing activity, similar to automatic writing behavior.

Case Report

The patient was a 79-year-old, right-handed woman who could independently perform daily living activities, including cooking, washing, and cleaning. One day she suddenly lost spontaneous speech, responded to questions with a nod, and remained in bed all day. Two days later, she was admitted to our hospital for evaluation of decreased spontaneity. She had a history of angina, but had no history of psychiatric disease or cognitive impairment. The patient was alert, but indifferent to her surroundings. She showed no signs of motor paresis or sensory deficit. Snout and grasp reflexes were not observed. Focal dystonia was observed in her right hand. Blood levels of ammonia, thyroid hormone, and vitamin B₁₂ were normal. Brain magnetic resonance images (MRI) revealed ischemic lesions involving the inferior genu of the left internal capsule, a part of the internal segment of the globus pallidus, and subcortical white matter with mild cortical atrophy ([fig. 1A–C](#)). Diffusion-weighted MRI confirmed that the lesion in the left internal capsule of the genu extended to a part of the internal segment of the globus pallidus, and was responsible for the patient's symptoms ([fig. 1D](#)). Magnetic resonance angiography (MRA) showed no occlusions or stenoses in the main cerebral arteries. Technetium-99m ethyl cysteinate dimer single-photon emission computed tomography (SPECT) revealed a left-dominant diffuse hypoperfusion in the basal ganglia and frontal lobe ([fig. 1E, F](#)). Electroencephalography findings showed left-dominant theta activity of approximately 7 Hz, without epileptic discharges or triphasic waves.

Neuropsychological Findings

The patient was not depressed, but showed abulia and amnesia. She scored 7 points on the Mini-Mental State Examination scale (MMSE). She could follow simple commands without speaking. There was neither apraxia nor agnosia. Visual-spatial testing was normal. The Wechsler Adult Intelligence Scale-Revised and the Trail Making Test were discontinued halfway through the assessment due to decreased spontaneity. Verbal comprehension and naming were mildly impaired, while repetition and reading were preserved. There was no verbal perseveration. Her writing was severely impaired; she could spell only a few words correctly, although when asked to write her name, she exhibited neologism in kanji and wrote 'Ishimura-shinbun', combining 'Ishimura' (her last name) and 'shinbun' (the newspaper placed before her) ([fig. 2A](#)). When directions to write her favorite food, hometown, and current date or place were given, she continued to write neologistic kanji words related to her last name. However, after the word 'shinkeinaika' (neurology) was placed in front of her as a written note, she wrote the same word in place of her name and continued writing. The writing was mainly perseverative with lexicographic alterations; many words written along the lines were legible and without spatial distortions. Increased writing activity was observed when ruled paper and a pen were presented to her; however, this was not observed when blank paper was presented. There was no frustration or anxiety when the pen was removed. She could use other tools such as a toothbrush, scissors, and a comb accurately; utilization behavior was not elicited by presenting those items. She also showed perseveration in drawing figures ([fig. 2B](#)). On day 21, when she was asked to write something (her name, her daughter's name, current date or her feelings), she wrote only a few words related to her last name; her increased writing characteristic disappeared completely, except for a slight perseveration. Two years later, her response time to questions had become slightly shorter; however, she replied in a small voice, with a smile, and continued to exhibit decreased spontaneity.

Discussion

This is the first report of an increased writing activity, together with perseverative writing, that was related to a capsular genu infarction. However, verbal perseveration in patients with capsular genu infarctions has been reported previously, and all patients had left-sided infarctions and showed abulia ([table 1](#)) [5–8]. Lai et al. [9] reported a 46-year-old patient with a left capsular genu infarction who exhibited auditory hallucinations and dysgraphia, and whose symptoms predominantly included paraphasia and character error without increased writing activity or perseveration.

Van Vugt et al. [10] reported a 70-year-old right-handed man with a history of alcohol abuse who demonstrated progressive memory disturbance (score of 11 points on the MMSE), verbal asponaneity, and increased writing activity, which was described as

‘automatic writing behavior’. This diagnosis was based on the perseverative, legible writing pieces of language that he produced both visually and verbally at the syntactic or lexical level, and well-ordered spatial arrangement. On the basis of SPECT findings from right-dominant frontal hypoperfusion, the mechanism was considered to be a particular form of utilization behavior caused by suppression of the inhibitory system of the frontal lobes. In contrast, hypergraphia reportedly comprises large numbers of sentences with copious unrepeated content. The written characters are difficult to read and spatial placement is not ordered [1]. The increased writing activity seen in our patient was more similar to automatic writing behavior than hypergraphia.

Evyapan and Kumral [11] studied automatic writing behavior in three patients with right cerebral hemispheric strokes. They suggested that a combination of visuospatial neglect and frontal dysfunction may be responsible for the mechanism, as all of the patients showed left visuospatial neglect, and writing behavior was elicited only by the visual letters on the right side. In contrast, the neologism in our case of ‘Ishimura-shinbun’ (see fig. 2A) is considered to represent automatic reproduction of the visual stimulus, although our patient did not show visuospatial neglect. In the study by van Vugt et al. [10], the duration of the patient’s automatic writing behavior was permanent, while our patient’s writing characteristic was temporary and similar to that reported by Evyapan and Kumral [11].

Automatic writing behavior in our patient manifested after the oral or written direction to write, or the presence of ruled paper and a pen. It was probably due to a form of motor perseveration related to the basal ganglia. The pallidum and substantia nigra are the sites of input from motor corticostriatal pathways that interact as part of normal motor control and the termination of movement [12]. Four patients with infarction in the thalamus and the basal ganglia developed motor perseveration of the upper and lower extremities induced by passive movement. One of these patients demonstrated perseveration in drawing figures, which suggests impairment of the prefrontal cortico-basal ganglia-thalamocortical loops [13]. In this case, SPECT images revealed diffuse hypoperfusion in the left frontal lobe and basal ganglia, which implies that damage in the anterior and inferior thalamic peduncles, adjacent to the capsular genu, led to thalamocortical disconnection [8] and interruption of the cortico-basal ganglia-thalamocortical loops [14].

Our patient’s repeated writing of her last name may indicate dysfunction of recall and understanding of characters in the dominant hemisphere, which is a requirement for writing [15]. Moreover, a previous study reported that more severe verbal memory loss is caused by lesions on the left side of the capsular genu, compared with lesions on the right [8]. As described in a previous report [11], the transient quality of automatic writing behavior implies possible diaschisis. It remains unclear why other perseverative movements, in addition to writing, did not occur. However, with regard to speech, we assume that the absence of verbal perseveration can partially be attributed to the effect of severe abulia.

Our patient showed focal dystonia in her right hand. We suggest that this may be due to the involvement of a part of the left internal segment of the globus pallidus. Focal dystonia has been thought to be responsible for globus pallidus and thalamic infarction [16].

In our case, the main blood supply route to the capsular genu was undetermined, since cerebral angiography was not performed. However, based on MRA results, our patient did not have occlusions or severe stenoses in the main arteries. It has been reported that

the capsular genu is supplied by perforating arteries, arising from the apex of the internal carotid or anterior cerebral artery, or by the anterior choroidal artery [17].

The patient was not examined for cognitive or mental function before the onset of the capsular genu infarct; however, we believe that she did not have dementia or psychiatric diseases on admission, since her family attested that before the stroke she had been able to perform her usual activities properly and had showed no personality changes. Moreover, two years after the stroke, the patient showed persistent abulia that did not progress further. Therefore, we suggest that the acute-onset abulia and automatic writing behavior were likely due to the new infarct in the capsular genu (corresponding to high signal intensity on the diffusion-weighted images) rather than concomitant psychiatric or neurodegenerative disorders that exhibited dementia. However, we cannot exclude the possibility that an interaction between a concomitant underlying cognitive dysfunction, due to mild cortical atrophy or subcortical multiple infarcts, and the capsular genu infarct may exist. This may then cause the characteristic writing behavior, since leukoaraiosis has been associated with subtle cognitive impairment [18].

In conclusion, we suggest that malfunction of the frontal-subcortical circuit, due to an inferior capsular genu infarction, contributed to the development of motor perseveration in writing.

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Table 1. Review of perseveration in patients with capsular genu infarction

Author	Year	Age/ gender	Handed- ness	Lesion site	Abulia	Amnesia	Perseveration
Tatemichi et al. [8]	1992	60/M	R	left capsular genu	+	+	verbal perseveration
Moreaud et al. [7]	1996	23/M	R	left capsular genu	+	–	verbal perseveration
Chukwudelunzu et al. [5]	2001	71/F	R	left capsular genu	+	+	verbal perseveration
Hayashi et al. [6]	2003	72/F	R	left capsular genu and left thalamus	+	+	verbal perseveration
Present case	2009	79/F	R	left capsular genu	+	+	perseveration in writing

Fig. 1. **A, B** T2-weighted axial and coronal magnetic resonance images (MRI; TR/TE, 4,000/100) show that the high-signal lesion involves the left inferior genu and part of the internal segment of the globus pallidus (arrows). **C** Fluid attenuated inversion recovery images (TR/TE, 8,000/120) showing periventricular hyperintensities (arrowheads) and multiple lacunar infarctions (arrows). Mild cortical atrophy was noted. **D** Diffusion weighted MRI (TR/TE, 2,500/82) showing a recent lesion in the genu of the left internal capsule extending to a part of the internal segment of the globus pallidus. **E, F** Technetium-99m ethyl cysteinate dimer (^{99m}Tc -ECD) single-photon emission computed tomography (SPECT) images show diffuse hypoperfusion in the left frontal lobe and basal ganglia (arrows).

Fig. 2. A The patient continued to write a neologistic kanji (morphograms) word, 'Ishimura-shinbun', which combines 'Ishimura' (her last name) and 'shinbun', which is the name of the newspaper that had been placed before her. Although an additional request was made (to write her birthday and the date at that time), she continued to write similar kanji words, with the following lexicographemic alterations: 'Ishimura-shinbun', 'Ishimura-shinichi', and 'Ishimura-buichi' (arrowhead). However, after the word 'shinkeinaika' (neurology) was placed in front of her in the form of a note (see arrow), the patient wrote 'shinkeinaika' in place of her name and continued writing. **B** When the figure was placed in front of her in the form of a note (see arrow), the patient continued to draw the same figure.

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