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Case report

Agraphia of the left hand with dysfunction of the left superior parietal region without callosal lesions

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ABSTRACT

A 28-year-old right-handed man noticed weakness in his legs, three days after an ephedrine overdose. Initial brain magnetic resonance imaging showed lesions in the parietal regions bilaterally. Computed tomography angiography showed segmental and multifocal vasoconstriction of the cerebral arteries. After treatment, clinical and radiological findings resolved, suggesting the patient had reversible cerebral vasoconstriction syndrome with posterior reversible encephalopathy syndrome. However, he had residual agraphia of the left hand. Language testing revealed no difficulties in oral expression, auditory comprehension, understanding of written language, or writing with the right hand. I-123 iodoamphetamine single-photon emission computed tomography showed residual dysfunction in the left superior parietal lobule. There were no apparent signs of other disconnection syndromes or neuroimaging abnormalities in the corpus callosum. We diagnosed left-hand agraphia due to left parietal dysfunction. Our case suggests that left superior parietal dysfunction without callosal lesions is a possible cause of left-hand agraphia. Neural mechanisms for writing with the right or left hand may be separable at the cortical level.

1. Introduction

Agraphia of the left hand is a classical disconnection syndrome [1]. The functional explanation is that right hemispheric cortical representation of the left hand gets deprived of access to linguistic information represented in the left hemisphere. Normally, transfer of such information relies on integrity of the anterior commissural fibers traveling through the corpus callosum. Therefore, the structural reason for such a finding is often a callosal lesion. We report a case of left-hand agraphia with selective dysfunction in the left superior parietal region without callosal lesions.

2. Case report

A 28-year-old right-handed man suddenly noticed weakness in his legs, three days after an ephedrine overdose. There was no history of language disorders. On admission, he showed paraplegia, but otherwise unremarkable. His laboratory results were normal, including vitamin B1, folic acid, thyroid hormone, angiotensin-converting enzyme, lactate acid, and pyruvate acid levels. Hepatitis B and C virus screening were negative. Cerebrospinal fluid examination showed normal cell count (1/μL), glucose (57 mg/dL), and protein levels (42 mg/dL).

Initial brain magnetic resonance imaging (MRI) showed multiple symmetric hyperintense areas in the left occipital and both parietal lobes on both T2-weighted imaging and diffusion-weighted images (DWI), corresponding to areas of increased proton diffusion on apparent diffusion coefficient (ADC) maps, indicating the presence of vasogenic edema (Fig. 1A). Furthermore, some lesions were hyperintense on DWI but had decreased proton diffusion on ADC maps, indicating the co-existence of cytotoxic edema. Computed tomography angiography of the brain showed multiple irregular, narrowed segments of multiple branches of both internal carotid arteries. Clinical and radiological abnormalities rapidly improved after oral calcium-channel blockers and cilostazol (Fig. 1B). Follow-up MRI showed residual partial hyperintense areas only in the superior parietal gyri bilaterally. Based on these clinicoradiological features, we diagnosed a cerebral infarction associated with reversible cerebral vasoconstriction syndrome and posterior reversible encephalopathy syndrome due to ephedrine overdose [2].

On follow-up neuropsychological assessment, the patient had difficulty writing with the left hand. He showed no apparent signs of other disconnection syndromes. The screening test for aphasia revealed no apparent difficulties in oral expression, auditory comprehension, or understanding of written language. Writing with the right hand was normal.

To further assess his left-hand agraphia, we conducted a writing test

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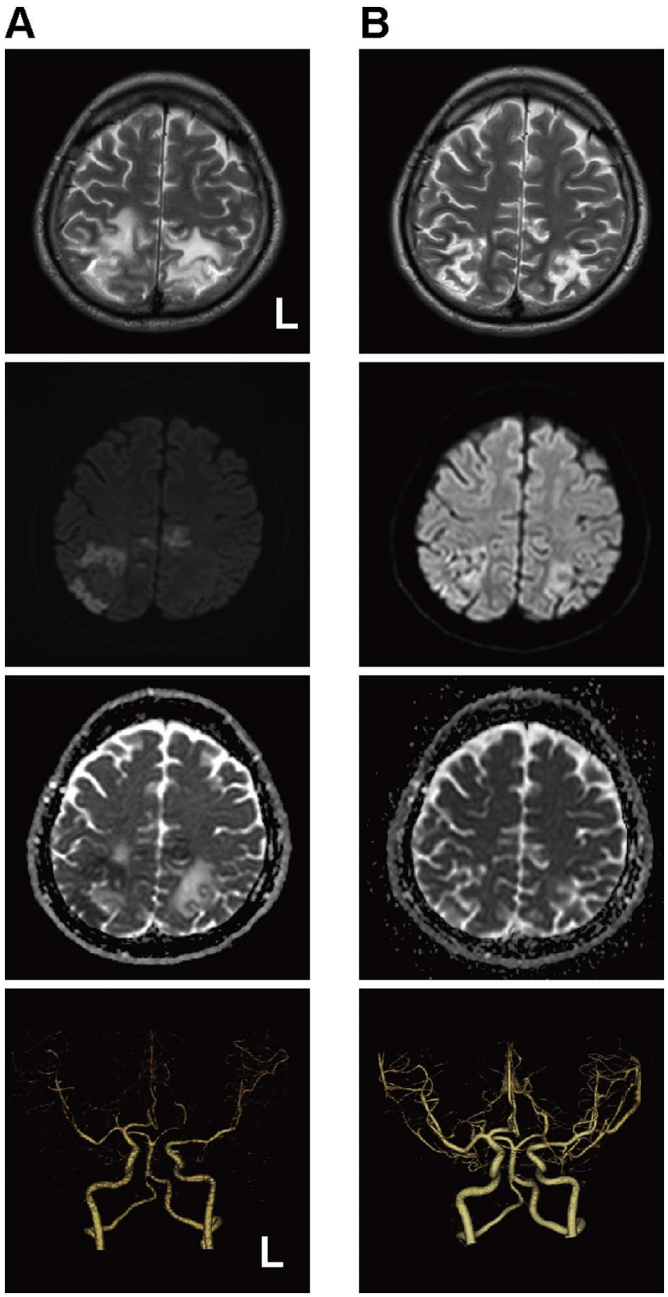

Fig. 1. Neuroimaging findings in a patient with left-hand agraphia. (A) Day 1 MRI and 3D-computed tomography angiography (CTA) findings. The right, right middle, left middle, and left figures are T2-weighted and diffusion weighted images, apparent diffusion coefficient maps, and 3D-CTA, respectively. (B) Follow-up MRI (day 27) and 3D-CTA (day 16). Abnormalities in the initial neuroimaging study were improved, suggesting reversible cerebral vasoconstriction syndrome with posterior reversible encephalopathy syndrome.

involving words he could write before this illness (Fig. 2). The Japanese language consists of two writing systems: kanji (morphograms or ideograms) and kana letters (syllabograms, including simpler phonograms) [3]. We assessed the patient's ability to write kanji and kana separately. His left-hand agraphia was characterized by impaired kanji and kana character recall, poor grapheme formation, and incorrect stroke sequences. This kind of trial and error was observed only when writing with the left hand. When copying a word, stroke sequences were incorrect. He could write symbols such as circles, squares, and triangles without visual cues.

Residual MRI lesions included areas in superior parietal lobules,

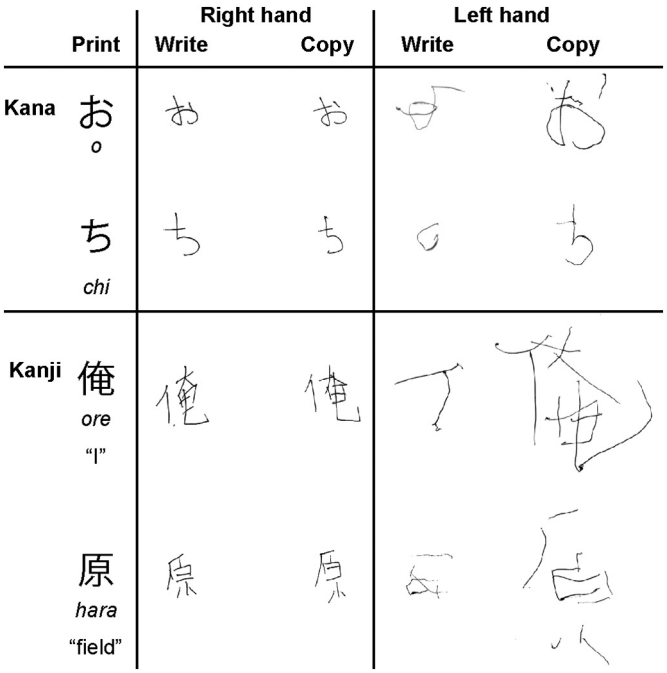

Fig. 2. Writing samples. Samples of writing and copying with left and right hand are shown. Copying the character with the left hand was better than writing the character on command.

intraparietal sulcus, and the left supramarginal gyrus. There were no apparent abnormalities in the corpus callosum (Fig. 3A). I-123 iodoamphetamine single-photon emission computed tomography showed prominent hypoperfusion in the left superior parietal gyrus (Fig. 3B). Considering these findings, the final diagnosis was left-hand agraphia after an ephedrine overdose due to dysfunction of the superior partial gyrus without callosal lesions.

3. Discussion

Our patient had residual lesions in the superior parietal lobules, the intraparietal sulcus, and the left supramarginal gyrus. Previous studies suggested that agraphia develops due to lesions of the left superior parietal regions [4]. We know of no reports on agraphia due to right superior parietal dysfunction. In our case, there was selective hypoperfusion in the left superior parietal region (Fig. 3B), although brain MRI showed bilateral superior parietal lesions (Fig. 3A). These findings indicate that left superior parietal dysfunction was responsible for his left-hand agraphia. One explanation for this observation is that neural mechanisms for writing with the right or left hand may be separable within the left superior parietal regions. This hypothesis is supported by another report of a patient with pure alexia (another of the disconnection syndromes [1]) but no callosal lesion [5]. It therefore appears that classical disconnection syndromes are possible even without a callosal lesion.

Neural mechanisms for agraphia for kana and kanji are considered to have both common and separate pathways [3]. Orthographic information goes from the posterior inferior temporal cortex (orthographic lexicon site) and proceeds upward under the angular gyrus and superior parietal lobule to travel to the frontal motor and premotor area (orthographic route), whereas phonological information goes from the posterior superior temporal gyrus (phonological lexicon site) to the angular and supramarginal gyri and joins the arcuate fasciculus to travel to the frontal motor and premotor area (phonological route) [4]. These two lexicons have a reciprocal connection. According to this hypothesis, graphically complex kanji characters depend more on the orthographic route, with kanji agraphia resulting from damage to any

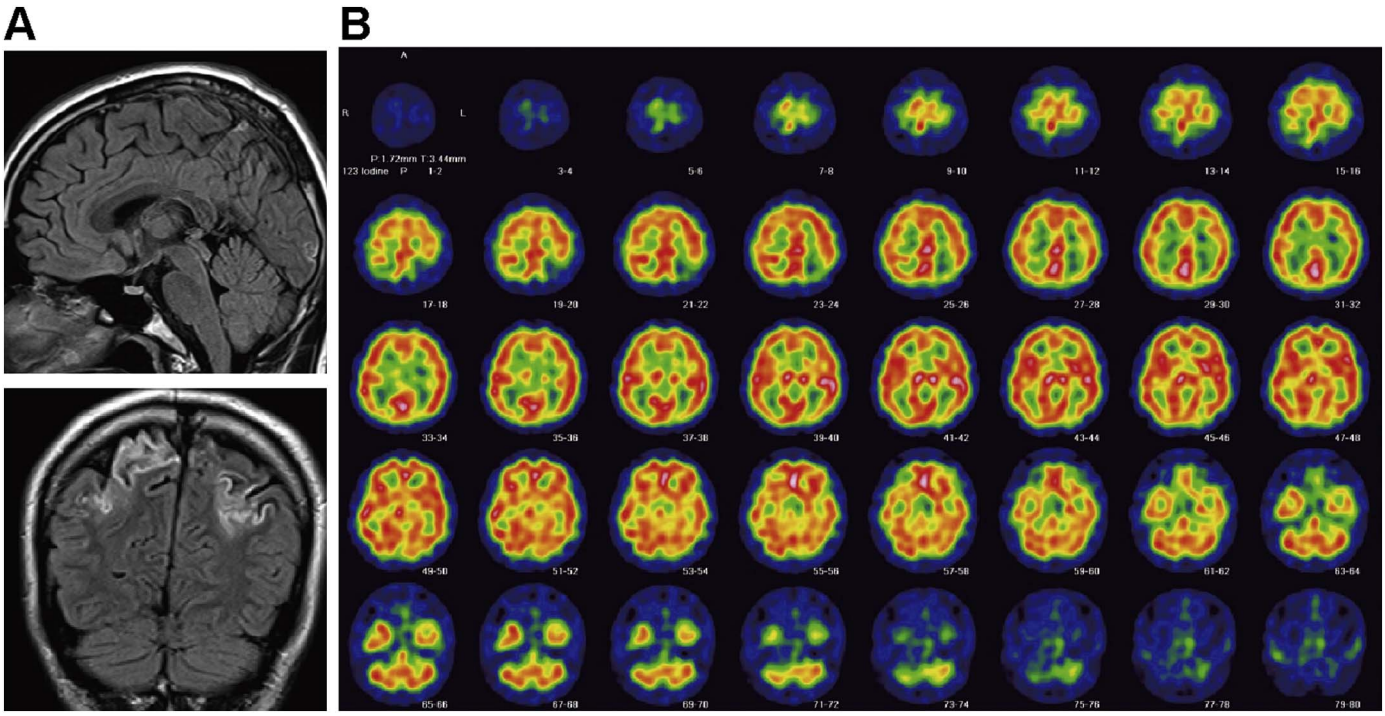

Fig. 3. Neuroimaging findings associated within left-hand agraphia. (A) Sagittal and coronal fluid attenuated inversion recovery images showing no callosal lesions. (B) Single-photon emission computed tomography showing selective hypoperfusion in the left superior parietal region.

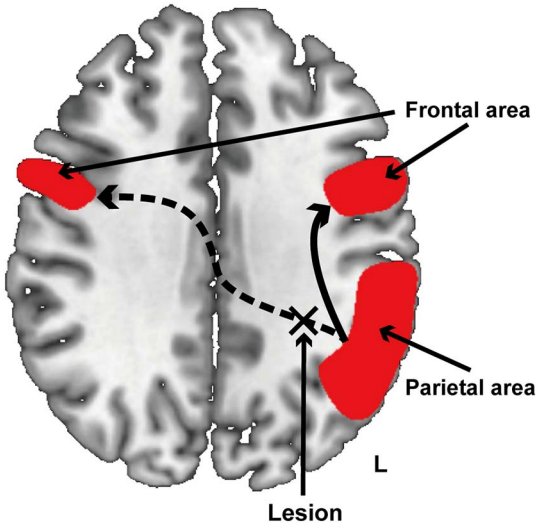

Fig. 4. Schematic representation of the possible mechanism for left hand agraphia due to the left superior parietal lesion. The connection between the right frontal area and the left parietal area (a curved dotted line) is supposed to be disrupted by the subcortical lesion in the left parietal area.

part of that route. In contrast, graphically simple kana characters are directly linked to phonemes and depend less on the orthographic route; hence, kana agraphia occurs less frequently with orthographic route lesions. Our patient had agraphia for both kanji and kana (Fig. 2), suggesting the possibility that both orthographic and phonological routes were disrupted. The basic idea is that the left-hand agraphia could result from commissural disconnection [1]. Therefore, it is possible that the subcortical lesion within the left superior parietal regions induces the disconnection between the right frontal motor and premotor area and the left parietal area for our patient (Fig. 4). The unilaterality of his agraphia indicates that such neural connections for the left and right hands are separable within the left parietal regions.

4. Conclusion

Our case suggested that dysfunction in the left superior parietal gyrus without callosal lesions is a possible cause for left-hand agraphia. Extensive assessment of writing ability would be important for rehabilitation and accurate diagnosis of cognitive factors.

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