



Case Reports

Micrographia after midbrain infarction alleviated by Japanese calligraphy-style writing: A case report

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Micrographia is a condition in which patients write abnormally small letters or symbols. This disorder presents in one of two forms: consistent micrographia, in which written letters are consistently small, and progressive micrographia, wherein initial letters are normal-sized and subsequent letters are progressively smaller. Although it is frequently observed in patients with Parkinson's disease, in rare cases, micrographia can occur following focal brain damage. The lesions responsible for micrographia occur in the frontal lobe, basal ganglia, and thalamomesencephalic region. In all cases reported to date, the lesions were located on the left side of the brain, with micrographia occurring in the right hand, except for one case reported by Lee [1], wherein micrographia was associated with a lesion localized to the midbrain tegmentum. In this case, the lesion was located on the right side, and micrographia occurred in the right hand. Micrographia is a frustrating condition for patients with brain damage or Parkinson's disease, and only a few treatment options are available.

East Asian calligraphy, a tradition unique to China, Korea, and Japan, is the art of expressing the beauty of written letters with a high level of artistic freedom using a brush. Herein, we describe the case of an individual with a lesion in the right midbrain who had micrographia in the right hand, which was resolved with Japanese calligraphy-style writing.

A 77-year-old right-handed man was admitted to our hospital after experiencing diplopia and difficulty walking. He practiced Japanese calligraphy as a hobby until 20 years before symptom onset. Neurologically, the patient presented with right medial longitudinal fasciculus syndrome, bilateral cerebellar ataxia, dysarthria, persecutory delusions, and frontal lobe dysfunction. Details of his delusions and frontal lobe dysfunction have been reported separately [2]. Progressive micrographia occurred in his right hand, but not in his left. Magnetic resonance imaging (MRI) revealed an infarct in the right midbrain, including the tegmentum (Fig. 1A).

The patient's delusions and frontal lobe dysfunction disappeared 4 months after onset [2], but his other neurological symptoms, including micrographia, remained. However, the patient discovered and implemented a compensatory approach for resolving micrographia. The patient expressed, "Letters look better when I write as though I am doing Japanese calligraphy." The patient's handwriting is shown in Fig. 1B. The mean height of the first five letters was 2.2 times that of the last five letters when writing in the usual manner (left); however, the mean height of the first five letters was only 0.9 times that of the last five when writing in a calligraphy-like manner (middle). The time required for writing a letter in the usual and calligraphy styles was 2.80 s and 2.65 s, respectively. Namely, although letters written in the usual manner

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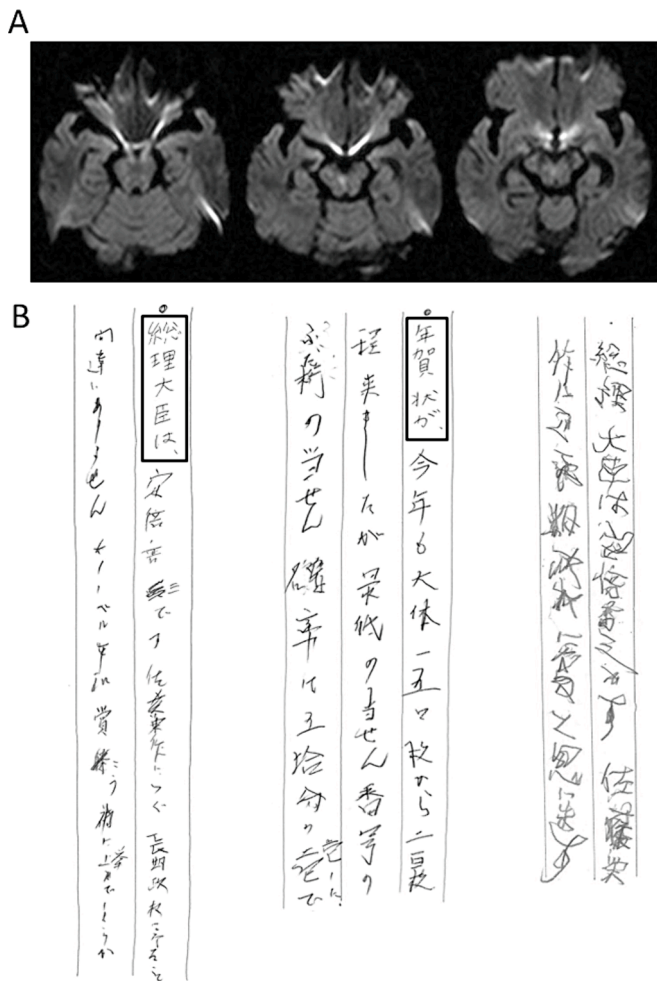


Fig. 1. Diffusion-weighted magnetic resonance imaging (A) and samples of handwriting (B) of the patient. (A) The infarct lesion was detected in the lower part of the midbrain below the red nuclei. It was on the right side of the medial midbrain tegmentum from the anterior to the posterior edges and in the right cerebral peduncle, including a part of the substantia nigra. No other lesions were detected. (B) The patient's usual handwriting (left), and calligraphy-like writing (middle) four months after the onset. The characters enclosed in squares have been written by the examiner. The right part of the figure shows the patient's handwriting with his left hand. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

continued to progressively decrease in size, those written in a calligraphy-like manner remained constant in size, even letters written using a pen instead of a brush. Although a brush and pen are generally held differently, the patient did not alter the way he held a pen when writing in a calligraphy-like manner. When asked about intentional differences between the two writing styles, he answered, "When I am writing, I recall how I felt during calligraphy writing."

This case study was approved by the ethics committee of Shinoda General Hospital, and written informed consent was obtained from the patient and his family in accordance with the Declaration of Helsinki.

In previous cases of micrographia caused by focal lesions, the lesions were located on the left side of the cortico-basal ganglia loop, and the symptoms occurred in the right hand. To the best of our knowledge, the present case and that reported by Lee [1], exclusively involving midbrain lesions, are the only known instances wherein micrographia in the right hand was associated with a right-side lesion. Although the basal ganglia and cerebellar loops have been assumed to be anatomically separate, research has revealed that they both have bidirectional

communication at the brain stem [3]. Lee [1] argued that the cause of his patient's micrographia may be the interruption of this communication. Moreover, a functional MRI (fMRI) study by Wu et al. [4] indicated that in patients with Parkinson's disease and progressive micrographia, functional interruption between the anterior part of the left supplementary motor area (SMA), which is a part of the cortico-basal ganglia loop, and the right cerebellum is observed in addition to the aberrant functioning of the cortico-basal ganglia loop. These findings suggest that in our case and Lee's case [1], the channel connecting the cortico-basal ganglia and cerebellar loops was interrupted owing to an injury in the midbrain tegmentum, causing micrographia.

Furthermore, Chen et al. [5] compared resting-state fMRI data between individuals who were proficient in Chinese calligraphy and those who were not. There were no differences in areas related to movement or sensory processing, such as the postcentral gyrus and SMA, but in the group proficient in Chinese calligraphy, the path lengths were shorter and the local efficiency was higher in the frontal lobe, parietal lobe, limbic system, caudate nucleus, and thalamus, suggesting superior organization in those areas. Similarly, in our report, writing in the style of Japanese calligraphy may have engaged these brain areas, thus compensating for micrographia.

In conclusion, this report suggests that Japanese calligraphy could be utilized as a therapy to improve micrographia.

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CRediT authorship contribution statement

Kazutaka Sakamoto: Conceptualization, Investigation, Methodology, Visualization, Writing – original draft. **Kyungshil Kim:** Writing – original draft. **Ryota Kobayashi:** Resources, Writing – review & editing. **Kayoko Yokoi:** Writing – review & editing. **Daichi Morioka:** Writing – review & editing. **Shinobu Kawakatsu:** Supervision, Writing – review & editing. **Akihito Suzuki:** Resources, Supervision. **Kazumi Hirayama:** Conceptualization, Methodology, Supervision, Writing – review & editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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