Case Report

Reversible cerebral vasoconstriction syndrome associated with a traditional Japanese training method under a waterfall named Takigyo: a case report

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Case: Takigyo is a traditional Japanese training method for psychosomatic conditions in which individuals meditate under a waterfall. A 55-year-old man presented with a mild headache and visual loss that occurred following Takigyo. On the day of admission, acute ischemic stroke was suspected based on brain magnetic resonance imaging examination. However, subsequent brain magnetic resonance imaging revealed reversible vasoconstriction of the cerebral diffuse segmental arteries.

Outcome: We diagnosed reversible cerebral vasoconstriction syndrome caused by Takigyo on the basis of his clinical course and image findings. He was treated with nifedipine and his clinical condition improved without recurrence.

Conclusion: We experienced a unique RCVS associated with Takigyo. It is important to accurately assess the etiology of headache with unusual circumstances and differential diagnosis including reversible cerebral vasoconstriction syndrome.

Key words: Headache, reversible cerebral vasoconstriction syndrome, Takigyo, vasoconstriction

INTRODUCTION

Reversible cerebral vasoconstriction syndrome (RCVS) is characterized by acute-onset, severe headaches, with or without additional neurological symptoms and signs, and diffuse segmental constriction of the cerebral arteries, with spontaneous resolution within 3 months.1–3 The usual clinical manifestation is an acute headache and self-limiting course without new symptoms after 1 month.4 Typically, a headache associated with RCVS is bilateral with posterior onset followed by diffuse pain, often accompanied by nausea, vomiting, photophobia, confusion, and blurred vision.5 Reversible cerebral vasoconstriction syndrome can occur spontaneously but can also be provoked by various precipitating factors, postpartum, or following exposure to various vasoactive substances including drugs, alcohol, medications, and blood products.2,3 A few hypotheses have been proposed that explain transient disturbances in the control of cerebral vascular tone, but the exact pathogenesis remains unclear.5 The syndrome is generally self-limiting and has a low incidence of recurrence.6 Most patients with RCVS experience complete resolution of headaches and angiographic abnormalities within days to weeks.6 We recently encountered a patient with RCVS associated with Takigyo. Takigyo is a traditional Japanese training method for psychosomatic conditions in which individuals meditate while sitting or standing under a waterfall. Some people use Takigyo as a religious ritual or ceremony, whereas others just want to experience the practice. Our search of published works failed to find any report on cardiovascular or cerebrovascular events associated with Takigyo.

CASE REPORT

A 55-year-old Japanese man with a history of anxiety disorder was admitted to our hospital because of moderate headache and visual loss. On the morning before admission in spring, he had visited a river in the mountains to experience Takigyo for the first time. He was very nervous before stepping into the river. Soon after stepping under the cold waterfall (with water falling from a height of approximately 3 m), he developed a moderate headache.
beginning at the back of his neck. He described his headache as reaching maximum intensity within 1 min, graded 6 out of 10 on a notional analogue severity scale. Although he stepped out of the waterfall after approximately 3 min, his headache persisted. Furthermore, he became aware of gradual visual loss. On the day of admission, he went to his primary care physician complaining of his unusual condition. Magnetic resonance imaging (MRI) of his brain suggested acute ischemic stroke, and he was referred to our hospital for further examination and treatment. He had a history of anxiety disorder treated with alprazolam 0.4 mg and sulpiride 150 mg daily. There was no prior history of head trauma, cerebral disabilities, or any vascular risk factors including diabetes, dyslipidemia, or hypertension. He had no allergies to any medications or food. He had smoked approximately 50–60 cigarettes a day for 35 years and drank alcohol occasionally.

On examination, his vital signs showed the following: body temperature, 36.1°C; blood pressure, 140/78 mmHg; heart rate, 78 b.p.m.; respiratory rate, 14 breaths/min; and oxygen saturation, 97% on ambient room air. Neurological examination showed double vision and homonymous left lower quadrantanopia. A stiff neck and the Kernig sign were absent. There were no abnormalities with sensation or strength throughout, with normal reflexes. Laboratory analysis of complete blood count, electrolyte and creatinine levels, liver function, and coagulation tests were normal. Results of testing for anticardiolipin antibodies, antinuclear bodies, and antineutrophil cytoplasmic antibodies were all negative. Cerebrospinal fluid examinations on day 2 revealed a protein level of 38 mg/dL, a glucose level of 63 mg/dL, and a leukocyte count of 9/μL. Electrocardiogram showed a normal sinus rhythm. A brain computed tomography scan showed no abnormality. Brain MRI studies were carried out on the day of admission at a previous clinic. Diffusion-weighted MRI (DWI), T2-weighted sequences, and fluid-attenuated inversion recovery (FLAIR) imaging showed slightly high signal intensity at a symmetrical lesion in the right occipital lobe under the cerebral cortex consistent with ischemic stroke (Fig. 1). Moreover, apparent diffusion coefficient (ADC)

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**Fig. 1.** Brain magnetic resonance imaging (MRI) and magnetic resonance angiography on the day of admission of a 55-year-old man with reversible cerebral vasoconstriction syndrome caused by Takigyo. A, Diffusion-weighted MRI. B, Apparent diffusion coefficient map. C, T2-weighted MRI. D, Fluid-attenuated inversion recovery (FLAIR) imaging shows a slightly high signal intensity at a symmetrical lesion in the right occipital lobe under the cerebral cortex (arrow head).
maps showed slightly high signal intensity in the same lesion. Magnetic resonance angiography (MRA) findings were almost normal, excluding the left persistent trigeminal artery. Because the initial presentation and investigations suggested acute ischemic stroke, we administered argatroban as antithrombotic therapy.

On the day after admission, follow-up brain MRI showed no abnormalities in the right occipital lobe under the cortex. However, MRA showed multifocal segmental stenosis in the bilateral anterior cerebral artery and posterior cerebral artery (PCA) (Fig. 2). Therefore, we suspected RCVS and treated with nifedipine 20 mg daily instead of argatroban.

On day 10 after admission, although his visual loss and headache had completely normalized, follow-up brain MRA revealed remarkable diffuse segmental stenosis in the bilateral middle cerebral arteries and bilateral PCA (Fig. 2). He was discharged on day 14 without recurrence and was followed up as an outpatient. Magnetic resonance angiography on day 46 showed improvement in the diffuse segmental cerebral arterial stenosis (Fig. 2). We diagnosed him with RCVS based on diagnostic criteria (Table 1). He continued to take nifedipine 20 mg daily to prevent vasoconstriction.

**DISCUSSION**

DiAGNOSTIC CRITERIA FOR RCVS have previously been proposed by experts\(^1\)\(^–\)\(^3\) and modified based on the results of studies.\(^7\) Although we considered causes of vasoconstriction leading to RCVS, Takigyo was strongly:

| **Table 1. Diagnostic criteria for reversible cerebral vasoconstriction syndrome** |
|---------------------------------------------------------------|
| Acute and severe acute headache (often thunderclap)           |
| with or without focal deficits or seizures                    |
| Uniphasic course without new symptoms more than              |
| 1 month after clinical onset                                  |
| Segmental vasoconstriction of cerebral arteries shown by      |
| indirect (e.g., magnetic resonance or computed tomography)   |
| or catheter direct angiography                                |
| No evidence of aneurysmal subarachnoid hemorrhage            |
| Normal or near-normal cerebrospinal fluid (protein concentrations <100 mg/dL, <15 white blood cells/µL) |
| Complete or substantial normalization of arteries shown by    |
| follow-up indirect or direct angiography within              |
| 12 weeks of clinical onset                                    |

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suspected. The observed radiological abnormalities in the present case were unique. On the day of admission, the occipital lobe changes under the cerebral cortex revealed by DWI, T2WI, and FLAIR were consistent with acute ischemic infarction. In contrast, slightly high intensity in the ADC map of the same lesions suggested vasogenic edema. The contradictory findings from these different imaging techniques revealed reversible changes within a few days. Brain edema is an early manifestation of RCVS, typically diagnosed within a few days of clinical onset, and it usually reverses entirely within 1 month of clinical onset, much earlier than vasoconstriction.3 Reversible brain edema occurs in 8–38% of all cases of RCVS.7 Although we considered RCVS as the most plausible differential diagnosis, posterior reversible encephalopathy syndrome (PRES) should also be considered because the pathological conditions of both RCVS and PRES are considered to be same.

There is a possibility to detect non-reversible lesions using single photon emission computed tomography, so it might have been an effective tool for differential diagnosis in this patient.

In this case, the patient might have experienced transient hypertension or rapid blood pressure changes that led to subsequent brain edema and cerebral vasoconstriction. We suggested that the observed clinical features, including visual defects, blurred vision, and photophobia, were caused by vasogenic brain edema in the occipital lobe and more distal vasoconstriction of PCA. Although the mechanisms of RCVS were unclear, the most plausible mechanism was the failure of the cerebral vascular autoregulation due to his severe nervousness and anxiety disorder. His vital signs were unknown just after Takigyo; however, his tension might have stimulated the sympathetic nervous system to suddenly raise his blood pressure. Considering his anxiety disorder, there might have been emotional changes associated with his health condition. As another factor, water falling from a height of approximately 3 m would have exerted some degree of external pressure on his head and neck. Some external impacts from Takigyo might have affected cerebral blood flow and led to vasoconstriction. There have been some cases of RCVS reportedly caused by head trauma.8,9 Although the patient described the waterfall speed as moderate and the weather was stable, the cold water in spring might have triggered his emotional changes and systemic vasoconstriction. His headache might have been associated with cold stimulus. Although we did not detect the definitive factor, there were several factors associated with his headache and RCVS. Finally, calcium channel blockers should be given as RCVS treatment, to prevent vasoconstriction. It is important to treat for cerebral infarction until it has been ruled out completely because discrimination between cerebral infarction, RCVS, and PRES is clinically difficult. When patients present with headache as in this situation, a differential diagnosis should be made according to the International Classification of Headache Disorders, 3rd version.10

CONCLUSION

We experienced a unique RCVS associated with Takigyo. It is important to accurately assess the etiology of headache with unusual circumstances and differential diagnosis including RCVS. This was a particularly rare case. Although many people experience Takigyo each season in Japan without any adverse effects, in our patient acute changes in emotion and a history of treated anxiety disorder could have been related to the cause of RCVS following Takigyo.

DISCLOSURE

Approval of the research protocol: N/A.
Informed consent: Informed consent was obtained from the patient for publication of this case report.
Registry and the registration no. of the study/trial: N/A.
Animal studies: N/A.
Conflict of interest: None declared.

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