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Recurring cervical internal carotid artery vasospasm elicited by head rotation: illustrative case

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BACKGROUND Idiopathic extracranial internal carotid artery vasospasm (IEICAV) is a rare cerebrovascular disorder causing transient or permanent cerebral ischemia. The pathophysiology underlying this disorder is not well understood. Although various medical treatments have been tried, complete remission of vasospasm is difficult to achieve. The first case of bilateral IEICAV induced by head rotation, which was successfully treated by carotid artery stenting (CAS), was presented.

OBSERVATIONS A 40-year-old woman with bilateral IEICAV had been conservatively treated for 13 years. However, transient ischemic attacks (TIAs) were not suppressed. She eventually presented with a large brain infarction in the left hemisphere and was referred to our department. Digital subtraction angiography clearly demonstrated the triggering of internal carotid artery (ICA) vasospasm by head rotation. After CAS treatment, the TIAs disappeared completely.

LESSONS Clinicians should recognize that ordinary mechanical stimulation to the ICA by head rotation can induce vasospasm, and CAS should be performed for refractory IEICAV at the appropriate time to avoid cerebral infarction causing severe neurological deficits.

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KEYWORDS idiopathic extracranial internal carotid artery vasospasm; carotid artery stenting; cerebral infarction; stroke

Idiopathic extracranial internal carotid artery vasospasm (IEICAV) causing recurrent paroxysmal vasospasm is rare. Although migraine, smoking, and the cold pressor test have been implicated in such vasospasm, the definitive mechanism remains obscure, and the relevance of head rotation to the vasospasm has not been reported.[1–5] Medications such as calcium channel blockers and antithrombotic drugs have generally been used. However, complete prevention of the vasospasm seems difficult, occasionally leading to brain infarction.[6] Carotid artery stenting (CAS) has been performed for such refractory cases, but its effectiveness, including long-term outcomes, has not been established.[6–8] Herein, we report the first case of bilateral IEICAV that was induced by head rotation and successfully treated with CAS.

Illustrative Case

A 40-year-old, nonsmoking woman without any significant medical history presented with transient aphasia and right hemiplegia with a left cervical bruit 13 years earlier. Computed tomography angiography showed transient narrowing of the left internal carotid artery (ICA) at the first to second cervical vertebral levels. The transient ischemic attack (TIA) and narrowing of the ICA on imaging disappeared within at least a few days each time. Her symptoms then recurred repeatedly. From 2 years after the initial attack, they happened bilaterally. She was diagnosed as having bilateral IEICAV and received oral medication (verapamil, romeridine, aspirin, valproic acid, prostaglandin E1, nicorandil, isosorbide dinitrate, edoxaban). However, over the 13-year period, she had 63 episodes of

ABBREVIATIONS CAS = carotid artery stenting; DSA = digital subtraction angiography; ICA = internal carotid artery; IEICAV = idiopathic extracranial internal carotid artery vasospasm; TIA = transient ischemic attack.

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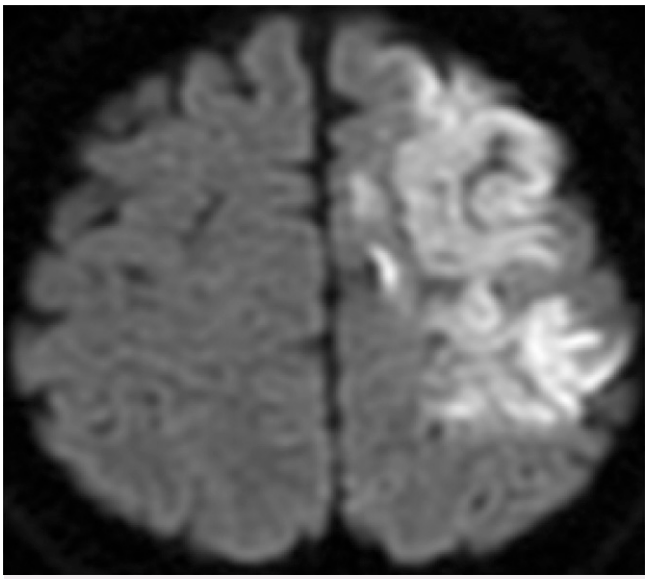

FIG. 1. Diffusion-weighted magnetic resonance imaging revealed cerebral infarction in the left cerebral hemisphere.

TIA with a cervical bruit (left side: 52 times; right side: 11 times). The frequency of TIAs ranged from 1 to 12 times per year.

In the 13th year, she had a large cerebral infarction of the left hemisphere resulting in aphasia and right hemiplegia (Fig. 1) and was referred to our department to prevent further cerebral infarction. To rule out Eagle syndrome, dynamic digital subtraction angiography (DSA) was performed during head rotation. Her styloid processes were not long enough to compress the bilateral ICAs during head rotation. However, right ICA vasospasm was unexpectedly induced by head rotation to the left side (Fig. 2A and B). The vasospasm worsened during the initial 10 minutes (Fig. 2C) but was relieved gradually and almost disappeared 25 minutes after onset (Fig. 2D). Based on these clinical findings, it was decided to perform bilateral CAS to prevent further ischemic stroke. Dual antiplatelet therapy with aspirin 100 mg/day and clopidogrel 75 mg/day was started 2 weeks before CAS. Right CAS was performed using a Carotid Wallstent 8 $\times$ 21 mm (Stryker Neurovascular), followed by left CAS using a Carotid Wallstent 8 $\times$ 29 mm 2 weeks later. After CAS, TIAs and cervical bruits on both sides disappeared completely for more than 2 years.

Discussion

Observations

The present patient had repeated episodes of vasospasm for many years, resulting in severe cerebral infarction. It was possible to induce vasospasm by head rotation as noted on DSA. This finding suggested that an ordinary body movement such as head rotation could induce vasospasm.

IEICAV is a rare disease, and only clinical case reports have been available. The clinical details, including etiology, symptom duration and time of onset, and effective treatment, are not yet fully understood.

There have been 22 reported cases of IEICAV, which are summarized in Table 1.[1–20] The mean age was 31.4 years (16–67), and females were predominantly affected (72.7% vs 27.3%). There were

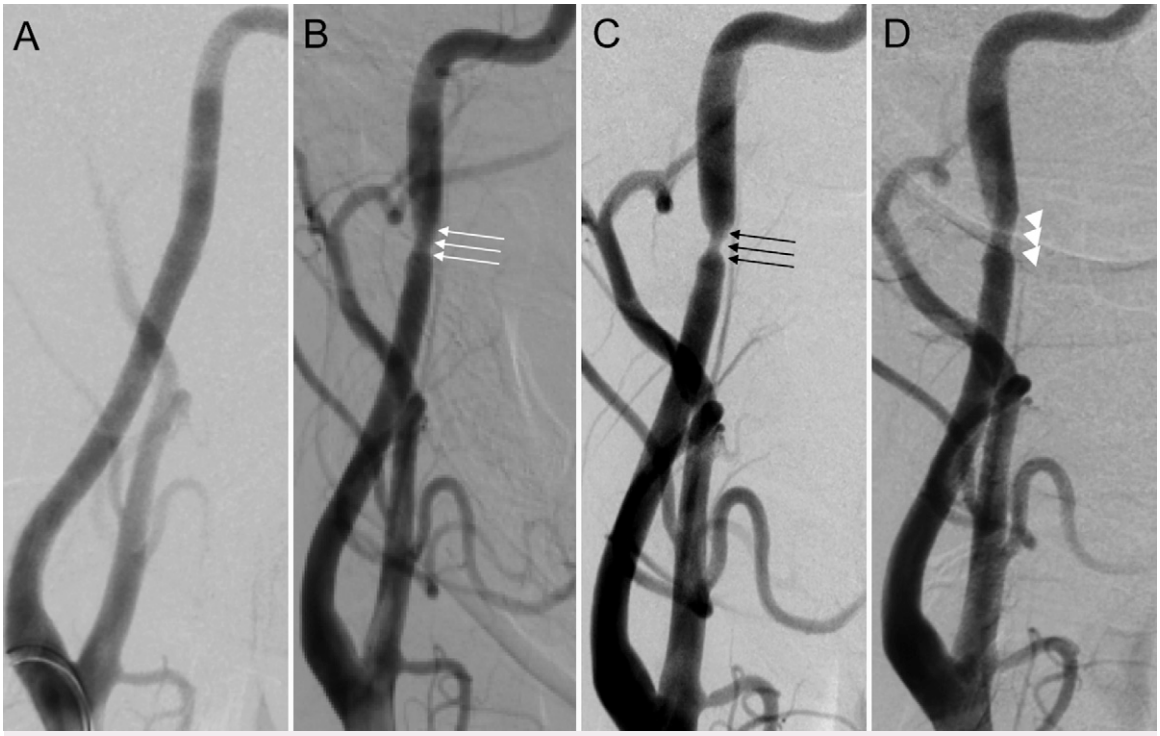

FIG. 2. Right common carotid artery angiography. **A:** ICA vasospasm was not seen on the initial angiogram. **B:** Vasospasm was induced immediately after head rotation (*white arrows*). **C:** Vasospasm was most exacerbated 10 minutes after head rotation (*black arrows*). **D:** Vasospasm was gradually improved (*white arrowheads*).

TABLE 1. Characteristics of 22 patients with carotid artery vasospasm

Variable	Value
Mean age in yrs (range)	31.4 (16–67)
Sex	
Male	6
Female	16
Side of vasospasm	
Right	4
Left	9
Both	9
Trigger	
Yes*	5
No	17
Cerebral infarction	10
mRS score	
0 or 1	19
NA	3
Median FU period in mos (range)	19 (6–48)
Medication	
Anticoagulant	6
Antiplatelet	16
Calcium blocker	13
Steroid	6
αβ blocker	2
β blocker	1
Magnesium	1
Nitroglycerin	1
Acetazolamide	1
Selective Serotonin Reuptake Inhibitor	2
Endovascular treatment	
CAS	6
Other†	3
Frequency of symptom after therapy	
Medication only	
Disappearance	5
Decrease	4
No change	3
NA	4
Endovascular treatment	
Disappearance	4
NA	2

FU = follow-up; IA = intraarterial injection; mRS = modified ranking scale; NA = not assessed; PTA = percutaneous transluminal angioplasty.

* Migraine = 3; smoking = 1; cold stimulation = 1.

† PTA = 2; calcium blocker IA = 1.

9 cases on both sides (40.9%), 9 cases on the left side (40.9%), and 4 cases on the right side (18.2%). Cerebral infarction was observed in 10 cases. Excellent clinical outcomes (modified Rankin Scale score 0–1) were achieved in 19 patients (95%), except for the present case (modified Rankin Scale score 3). With oral drug treatment alone, paroxysmal attacks disappeared in 5 cases (42%), decreased in 4 cases (33%), and were unchanged in 3 cases (25%). The most common oral treatment involved antithrombotic drugs (19 cases: anticoagulants 6, antiplatelets 16), followed by calcium channel blockers (13 cases). Nine endovascular treatments were performed in 6 cases. Four emergency interventions (intraarterial injection of vasodilator: 1 case; percutaneous transluminal angioplasty: 2 cases; CAS: 1 case) and 5 scheduled CAS procedures were performed.[4–7,11] Vasospasm improved in all patients, and there was no recurrence of vasospasm after CAS.

In 5 of the 22 reported cases, the triggers of IEICAV were suspected to be migraine in 3 cases, the cold pressor test in 1 case, and smoking in 1 case.[1–5] Patients with migraine may have genetic endothelial dysfunction,[21] which causes cerebral hypoperfusion by vasospasm of the extracranial ICA, as well as intracranial arteries, in the early phase of onset. The cold pressor test, which involves immersing the hand into an ice water container, activates the sympathetic nervous system and promotes the release of vasoconstrictive neurotransmitters such as noradrenaline and serotonin. This sympathetic stimulation may cause IEICAV through sympathetic innervation to the cervical ICA.[2] Smoking causes not only sympathetic stimulation via nicotinic receptors but also endothelial damage due to free radicals, which reduce nitric oxide release, and increase vasoconstrictors such as endothelin.[3] These suggest that patients with IEICV may have hypersensitivity to sympathetic nervous system stimulation. The present patient showed transient vasospasm after head rotation, which has never been previously reported. Therefore, the mechanism of IEICV induction by head rotation was not clear. However, the ICA at the C1–2 vertebral levels is well known to be the most vulnerable to mechanical stimulation by catheter or guidewire during cerebral angiography. It can also be hypothesized that patients with IEICAV may also have hypersensitivity to mild mechanical stimulation such as head rotation, which could induce vasospasm.

IEICAV has been reported to have a benign course. However, in some patients, oral medication alone did not completely prevent the paroxysmal attacks.[7] Because the present patient experienced severe brain infarction, oral medication alone carried a nonnegligible risk. On the other hand, CAS could prevent paroxysmal attacks completely without any perioperative complications.[6–8] Therefore, CAS should be performed in a timely manner when oral medication cannot effectively prevent symptomatic vasospasm.

Lessons

The first case of IEICAV induced by head rotation was reported. The patient experienced many repeated episodes of ICA vasospasm resulting in severe cerebral infarction. Cerebral angiography showed that vasospasm could be induced by head rotation, suggesting that an ordinary body movement such as head rotation can induce vasospasm in cases of IEICAV. CAS should be performed at the appropriate time for patients who are refractory to drug therapy.

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Kanemaru, Kaneko, Tateoka, Kinouchi. Acquisition of data: Kanemaru, Kaneko, Tateoka, Kinouchi. Analysis and interpretation of data: Kanemaru, Kaneko, Tateoka, Yoshioka, Wakai, Kinouchi. Drafting the article: Kanemaru, Kaneko, Tateoka, Wakai, Kinouchi. Critically revising the article: Kanemaru, Kaneko, Tateoka, Yoshioka, Hashimoto, Kinouchi. Reviewed submitted version of manuscript: Kanemaru, Kaneko, Tateoka, Kinouchi. Approved the final version of the manuscript on behalf of all authors: Kanemaru. Statistical analysis: Kaneko. Administrative/technical/material support: Kanemaru, Kaneko, Tateoka, Kinouchi. Study supervision: Kanemaru, Kaneko, Kinouchi.

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