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Idiopathic carotid and coronary vasospasm: A case treated by carotid artery stenting

Haruko Yoshimoto, Keizo Asakuno, Seigo Matsuo, Atsushi Ishida, Hideki Shiramizu, Kaku Niimura, Miki Yuzawa, Yasumichi Yamagishi[1], Takehiko Munakata[1], Takashi Moriyama, Tomokatsu Hori

Departments of Neurosurgery, [1]Cardiology, Moriyama Memorial Hospital (M.M.H.), 7-12-7 Nishikasai, Edogawa-ku, Tokyo 134-0088, Japan

E-mail: *Haruko Yoshimoto - yoshimoto@moriyamaikai.or.jp; Keizo Asakuno - asakuno-nsu@umin.ac.jp; Seigo Matsuo - sergio5679700@yahoo.co.jp; Atsushi Ishida - v2danyon@gmail.com; Hideki Shiramizu - h.shiramizu@gmail.com; Kaku Niimura - kaku4309@yahoo.co.jp; Miki Yuzawa - mellowseason@msn.com; Yasumichi Yamagishi - zvn05605@nifty.ne.jp; Takehiko Munakata - muna.muna.k1200gt@moriyamaikai.or.jp; Takashi Moriyama - kinenhp@moriyamaikai.or.jp; Tomokatsu Hori - thori@moriyamaikai.or.jp
*Corresponding author

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Abstract

Background: We previously reported a case of cerebral infarction complicated by myocardial infarction. The pathogenesis of both infarctions was thought to be vasospasm; thus, we named this condition 'idiopathic carotid and coronary vasospasm'. Various medical treatments for the prevention of carotid vasospasm have been unsuccessfully tried. Thus, other effective treatments should be established for patients who frequently suffer cerebral ischemic attacks.

Case Description: We treated the present case of 'idiopathic carotid and coronary vasospasm' by carotid artery stenting (CAS). The first stenting, of the carotid bifurcation, failed to prevent internal carotid artery (ICA) vasospasm. However, after an additional stent placement to the prepetrous portion, ischemic attacks were dramatically reduced.

Conclusion: The effect of CAS for extracranial ICA vasospasm was dramatic and control of the spasm at the prepetrous portion seems to be essential. Further validation of the effectiveness and safety of CAS for ICA vasospasm will be necessary.

Key Words: Carotid vasospasm, cerebral infarction, carotid stent placement, vasospastic angina, young patient

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INTRODUCTION

To date, several studies have reported transient and recurring stenosis of the extracranial internal carotid artery (ICA) [Table 1].[1,3-7,9-11,13,14] These cases usually showed completely resolved stenosis, to smooth and normal arterial walls, within hours to days, but had multiple recurrences. Thus, vasospasm has been presumed as the cause of the stenosis.

In 2011, we reported a case of a cerebral infarction complicated by myocardial infarction, both of which were caused by vasospasm. We named this condition 'idiopathic carotid and coronary vasospasm (ICCV)'.[14] Despite intensive medical treatment, this patient suffered repetitive transient ischemic attacks (TIAs). Four years after her first presentation, the patient developed prolonged aphasia and right hemiparesis because of the ICA vasospasm. Fujimoto *et al*. reported the successful

treatment of this patient by carotid artery stenting (CAS), the first in the literature.[4]

Shortly after that case, we encountered another example of this syndrome.

CASE REPORT

History and examination

A 40-year-old female without known cardiovascular risk factors or migraine episodes visited the Moriyama Memorial Hospital (M.M.H.) outpatient clinic in February 2011 after suddenly developing global aphasia and right hemiparesis. Emergent diffusion-weighted magnetic resonance imaging (DW-MRI) revealed fresh cerebral infarctions [Figure 1a, arrows]. Magnetic resonance angiography (MRA) revealed a suspicious stenotic lesion in the cervical segment of the left ICA [Figure 1b, arrow]. We performed digital subtraction angiography (DSA) 2 days after admission. No evidence of stenosis was found [Figure 1c, arrow]. MRA was performed again, and the result was the same as the DSA [Figure 1d, arrow]. Her laboratory data were negative for various coagulation disorders, collagen disease, and anticardiolipin syndrome. A Holter electrocardiogram (ECG) detected no arrhythmia; however, her ECG and cardiac echogram suggested an old myocardial infarction of the inferolateral wall, which was confirmed by myocardial scintigraphy [Figure 2a].

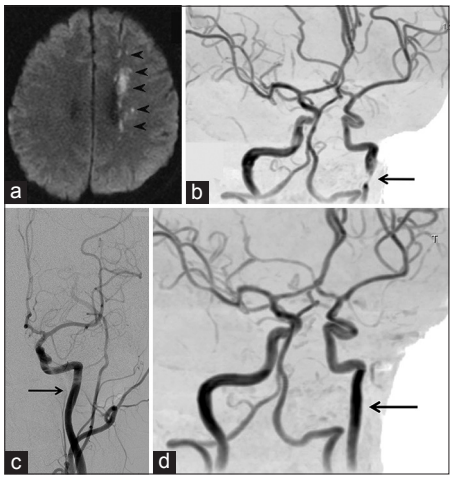

Figure 1: At onset of cerebral infarction, diffusion weighted magnetic resonance imaging (DW-MRI) revealed fresh infarctions in the left cerebral hemisphere (1-A, arrows). MRA revealed a stenotic lesion in the cervical segment of the left ICA (1-B, arrow). DSA (1-C, arrow), and MRA (1-D, arrows) on the second day after the onset revealed no evidence of stenosis

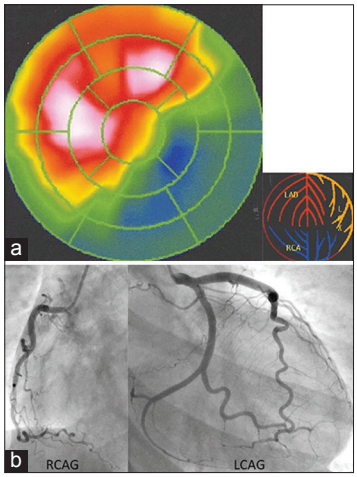

Figure 2: Myocardial scintigraphy identified the area without myocardial viability. Thereafter, an old myocardial infarction of the inferolateral wall was confirmed (Figure 2-a). Normal coronary angiography suggested that vasospasm was also the cause of the myocardial infarction in this case [Figure 2b]

Table 1: Extracranial ICA vasospasm in the literature

Author	Age	Sex	Age at onset	Heart disease	Migraine	Affected site
Lieberman (1984)[7]	39	Female	39	-	+	rt.→ blt. ICA, and rt.MCA
Rothrock (1988)[11]	31	Female	27	-	+	lt.→rt. ICA
Arning (1998)[1]	32	Female	32	Suspected vasospastic angina	-	blt. ICA
Kuzumoto (2005)[6]	31	Male	31	Atypical angina	-	rt. ICA
Janzarik (2006)[5]	30	Male	30	NS	-	rt.→ blt. ICA
Janzarik (2006)[5]	48	Female	48	-	+	rt.→ blt. ICA
Yokoyama (2006)[13]	35	Male	20	MI	NS	lt.→rt. ICA
Mosso (2007)[10]	45	Male	45	-	+	blt. ICA
Yoshimoto (2011)[14]	42	Female	39	MI due to vasospasm	-	lt.→ rt. ICA?
Moeller (2012)[9]	25	Male	12	NS	NS	blt. ICA
Dembo (2012)[3]	24	Female	24	Suspected angina	+	rt. ICA
Fujimoto (2013)[4]	47	Female	46	NS	NS	rt. ICA
This case (2014)	40	Female	29	MI due to vasospasm	-	lt. ICA

NS: Not stated, MI: Myocardial infarction, ICA: Internal carotid artery

Normal coronary angiography suggested that vasospasm was also the cause of the myocardial infarction in this case [Figure 2b]. With identical clinical features to our first case, we diagnosed this case as ICCV. In June and August 2011, she developed transient visual disturbance and right hemiparesis, and visited our hospital. ICA stenosis similar to the first presentation was detected by MRA, but disappeared in 2 days. Diltiazem hydrochloride and warfarin, which were continued from the first presentation, failed to prevent TIA.

Treatment and posttreatment course

The patient gave informed consent and CAS procedures were performed in accordance with our institutional guidelines in September 2011. Because the safety of stent deployment near to the first cervical vertebra, where the torsional stress might be larger than lower cervical levels, had not been established, CAS covering only the bifurcation in the same fashion as for atherosclerotic stenosis was performed, using a Carotid Wallstent® [Figure 3a]. However, symptoms suggesting vasospasm continued, and it presented twice radiologically during the 7 months following CAS [Figure 3b-d]. As seen in the figures, the vasospasm always occurs at just proximal portion of the petrous segment of ICA. Thus, in May 2012, we performed another CAS to cover the stenotic/spasm region of the ICA [Figure 3e]. Considering the possible torsional stress and the spasm strength of the carotid artery, we again used a Carotid Wallstent®, which is proven to have the highest external pressure among the stents available in Japan.[12] Since then, obvious cerebral ischemic attacks or ICA vasospasm have not been detected despite repeated MRA for a 24-month period following the additional CAS.

DISCUSSION

This is the second case we have reported for this rare condition 'ICCV' and its treatment with CAS.

Among 12 past patients, only 1 case almost identical to ours has been reported, by Kuzumoto *et al.*[6] That patient had no vascular risk factors or history of headache, but had a history of atypical angina.

In the literature, the focus has been on whether ICA vasospasm is related to migraine, like reversible cerebral vasoconstriction syndrome[2] as a migraine variant, or whether migraine headache is an epiphenomenon of vasospasm.[1,3-5,7,9,11,13,14]

Alternatively, it has been pointed out that the prevalence of migraine was significantly higher in the patients with vasospastic angina than in the control groups. On the basis of the result, Miller *et al.* proposed the concept 'generalized vasospastic disorder'.[8]

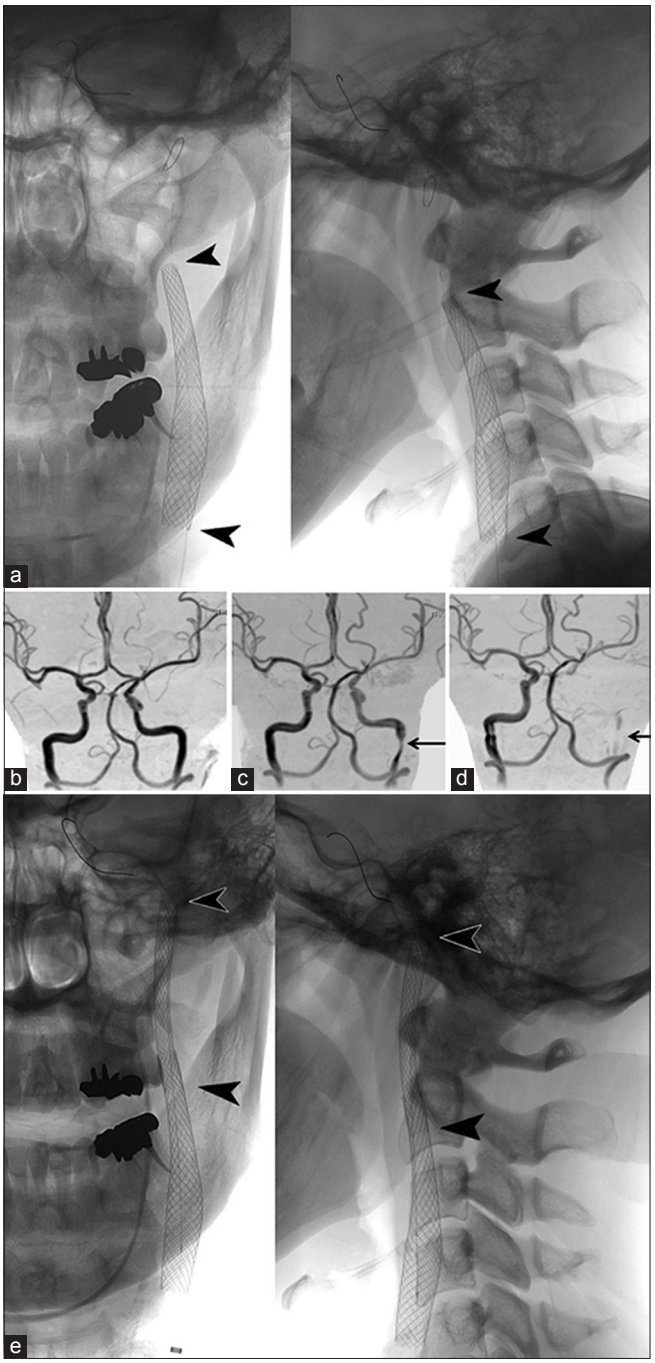

Figure 3: The first carotid stent was placed covering the bifurcation in the same fashion as for atherosclerotic stenosis from the level of the upper C2 superior endplate to the body of the C5 [Figure 3a, arrowhead]. Without vasospasm, MRA showed no blood flow defects, even after the first stenting [Figure 3b]. While the vasospasm was occurring, MRA revealed moderate [Figure 3c, arrow] or severe [Figure 3d, arrow] stenosis at the prepetrous portion of the internal carotid artery. The second stent was placed covering the stenotic/spasm region [Figure 3e, arrowhead]

If this concept was extended to include extracranial ICA vasospasm, it could consistently elucidate the combination of carotid vasospasm, migraine, and vasospastic angina in the past nine cases. However, it should be validated further.

Until now, few specific recommendations for the prophylaxis of extracranial ICA vasospasm could be made. Some papers report calcium antagonists[1,6,11] or α-blockers[9] could attenuate vasospastic changes and reduce symptom frequency. However, the effect has not proven consistent among past patients.[3,5,13,14]

CAS showed a curative effect for our previous patient.[4,14] On the other hand, our present case experienced a relapse at least twice after the first CAS. The difference of the initial CAS effect between the two cases introduced our next question: Is stenting to the prepetrous portion essential to subdue the spasticity of the ICA? Problematically, the safety of lifelong stent placement for younger patients at the high cervical vertebra level, with long-standing torsional stress, has not been established.

Our previous case experienced amaurosis fugax of the contralateral side proceeding to the initial side. Including this patient, in 9 (69%) of the past 13 patients, the affected side advanced from unilateral to bilateral, or both carotid arteries were initially affected. Patients presenting with ICCV may have a wide distribution of potentially sensitive arteries. Thus, they should be carefully followed and optimal medical strategies should remain sought.

Extracranial ICA stenosis is always relieved for hours to days. For this reason, diagnosis of extracranial ICA vasospasm is potentially difficult. Extracranial ICA vasospasm or ICCV should be included in the differential diagnoses for younger patients who suffered from cerebral infarctions of unknown etiology.

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