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

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

Departments of Neurosurgery, 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-nasu@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 - yzn05605@nifty.ne.jp; Takehiko Munakata - muna.muna.k1200@moriyamaikai.or.jp; Takashi Moriyama - kinenhp@moriyamaikai.or.jp; Tomokatsu Hori - thori@moriyamaikai.or.jp

*Corresponding author

Received: 10 April 14 Accepted: 21 August 14 Published: 30 October 14

This article may be cited as: Yoshimoto H, Asakuno K, Matsuo S, Ishida A, Shiramizu H, Niimura K, et al. Idiopathic carotid and coronary vasospasm: A case treated by carotid artery stenting. Surg Neurol Int 2014;5:S461-4.

Available FREE in open access from: http://www.surgicalneurologyint.com/text.asp?2014/5/13/461/143721

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

INTRODUCTION

To date, several studies have reported transient and recurring stenosis of the extracranial internal carotid artery (ICA) [Table 1].[1,2,7,8,11,13,14] 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.\(\text{[4]}\)

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].

---

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.\(\rightarrow\) blt. ICA, and rt.MCA |
| Rothrock (1988)\(^1\)   | 31  | Female | 27           | -             | +        | lt.\(\rightarrow\) rt. ICA |
| Aning (1998)\(^1\)      | 32  | Female | 32           | Suspected vasospastic angina | -        | blt. ICA |
| Kuzumoto (2005)\(^3\)   | 31  | Male  | 31           | Atypical angina | -        | rt. ICA |
| Janzarik (2006)\(^1\)   | 30  | Male  | 30           | NS            | -        | rt.\(\rightarrow\) blt. ICA |
| Janzarik (2006)\(^2\)   | 48  | Female | 48           | -             | +        | rt.\(\rightarrow\) blt. ICA |
| Yokoyama (2006)\(^1\)   | 35  | Male  | 20           | MI            | NS       | lt.\(\rightarrow\) rt. ICA |
| Mosso (2007)\(^1\)      | 45  | Male  | 45           | -             | +        | blt. ICA |
| Yoshimoto (2011)\(^1\)  | 42  | Female | 39           | MI due to vasospasm | -        | lt.\(\rightarrow\) rt. ICA |
| Moeller (2012)\(^4\)    | 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

---

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).
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. 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]

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\(^1,\text{6,11}\) or \(\alpha\)-blockers\(^9\) could attenuate vasospastic changes and reduce symptom frequency. However, the effect has not proven consistent among past patients.\(^3,\text{5,13,14}\)

CAS showed a curative effect for our previous patient\(^3,\text{4,14}\). 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.

REFERENCES

1. Arning C, Schrattenholzer A, Lachenmayer L. Cervical carotid artery vasospasms causing cerebral ischemia: Detection by immediate vascular ultrasonographic investigation. Stroke 1998;29:1063-6.
2. Calabrese LH, Dodick DW, Schwedt TJ, Singhal AB. Narrative review: Reversible cerebral vasospasm syndromes. Ann Intern Med 2007;146:34-44.
3. Dembo T, Tanahashi N. Recurring extracranial internal carotid artery vasospasm detected by intravascular ultrasound. Intern Med 2012;51:1249-53.
4. Fujimoto M, Isokawa H, Morita M, Okamoto N, Tomita Y, Kikuchi N, et al. Treatment of idiopathic cervical internal artery vasospasms with carotid artery stenting: A report of 2 cases. Journal of Neuroendovascular Therapy 2013;7:24-31.
5. Janzarik WG, Ringleb PA, Reinhard M, Rauer S. Recurrent extracranial carotid artery vasospasms: Report of 2 cases. Stroke 2006;37:2170-3.
6. Kuzumoto Y, Mitsui Y, Ueda H, Kusunoki S. Vasospastic cerebral infarction induced by smoking: A case report. No To Shinkei 2005;57:33-6.
7. Lieberman AN, Jonas S, Hass WK, Pinto R, Lin J, Leibowitz M, et al. Bilateral cervical carotid and intracranial vasospasm causing cerebral ischemia in a migraineur: A case of ‘diplegic migraine’. Headache 1984;24:245-8.
8. Miller D, Waters DD, Warnica W, Szlachcic J, Kreeft J, Theroux P. Is variant angina the coronary manifestation of a generalized vasospastic disorder? N Engl J Med 1981;304:763-6.
9. Moeller S, Hilt MJ, Blinzler C, Koehn J, Doerfler A, Schwab S, et al. Extracranial internal carotid artery vasospasm due to sympathetic dysfunction. Neurology 2012;78:1892-4.
10. Mosso M, Jung HH, Baumgartner RW. Recurrent spontaneous vasospasm of cervical carotid, ophthalmic and retinal arteries causing repeated retinal infarcts: A case report. Cerebrovasc Dis 2007;24:381-4.
11. Rothrock JF, Walicke P, Swenson MR, Lyden PD, Logan WR. Migraine stroke. Arch Neurrol 1988;45:63-7.
12. Wissgott C, Schmidt W, Behrens P, Brandt C, Schmitz KP, Andresen R. Experimental investigation of modern and established carotid stents. Rofo 2014;186:157-65.
13. Yokoyama H, Yoneda M, Abe M, Sakai T, Sagoh T, Adachi Y, et al. Internal carotid artery vasospasm syndrome: Demonstration by neuroimaging. J Neurol Neurosurg Psychiatry 2006;77:888-92.
14. Yoshihito H, Matsu S, Unemoto T, Kawakami N, Moriyama T. Idiopathic carotid and coronary vasospasm: A new syndrome?. J Neuroimaging 2011;21:273-6.