Recurrent Extracranial Carotid Artery Vasospasms
Report of 2 Cases

Wibke G. Janzarik, MD; Peter A. Ringleb, MD; Matthias Reinhard, MD; Sebastian Rauer, MD

Background and Purpose—Spontaneous vasospasms of the submandibular internal carotid arteries are rarely observed. They are a highly dynamic process, recur frequently, and can be detected by serial ultrasound examinations.

Summary of Cases—We present 2 cases of recurrent extracranial vasospasms of the internal carotid artery as a cause of stroke. In both cases, arterial dissection was initially suspected, but no intramural hematoma was detected on magnetic resonance imaging. Duplex sonography demonstrated recurrent high-grade stenoses of both internal carotid arteries that resolved spontaneously within hours to days. The vasospasms were treated with calcium antagonists and in 1 patient with oral corticoids.

Conclusions—Extracranial vasospasms as a cause of stroke might be underestimated. Vasospasms of the internal carotid arteries should be considered in patients with recurring ischemic events in the absence of any other explanation. Antiphlogistic treatment in combination with calcium antagonists might be effective to reduce the frequency of vasospasms. (Stroke. 2006;37:2170-2173.)

Key Words: internal carotid arteries ■ sonography ■ stroke, ischemic ■ vasospasm

Vasospasms of intracranial arteries are frequently observed as a complication of subarachnoid hemorrhage.1 Extracranial vasospasms are rarely reported and can be provoked by mechanical manipulation or drugs.2,3 Spontaneous submandibular vasospasms of the internal carotid arteries (ICAs) have been described in only a few cases,4–6 often associated with migraine. Ischemic stroke from reversible arterial vasospasms has been observed in a variety of conditions.7 Here we report 2 cases of recurrent vasospasms of the submandibular ICA as a cause of stroke.

Case Reports

Patient 1

A previously healthy 30-year-old man was admitted to the hospital with mild, left-sided brachiofacial hemiparesis and psychomotor retardation. Magnetic resonance imaging (MRI) showed an infarct in the anterior territory of the right middle cerebral artery. Duplex sonography detected an occlusion of the submandibular right ICA. Magnetic resonance angiography (MRA) and conventional angiography in combination with calcium antagonists might be effective to reduce the frequency of vasospasms. (Stroke. 2006;37:2170-2173.)

Key Words: internal carotid arteries ■ sonography ■ stroke, ischemic ■ vasospasm

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Duplex sonography demonstrated a high-grade stenosis of the right ICA. MRA a few days later revealed filiform stenoses of both submandibular ICAs. A duplex sonographic control 1 week later showed no stenosis of the right ICA but a high-grade stenosis of the left ICA. Bilateral dissection of the ICAs with underlying fibromuscular dysplasia was suspected, and treatment was started with intravenous heparin to maintain an activated partial thromboplastin time ratio of 2.0 to 2.5.

The patient had no cardiovascular risk factors, took no regular medication, and had no traumatic event during the preceding months. However, she had a history of severe bilateral pulsating headaches for at least 10 years. These episodes lasted between 2 and 5 days and had increased in frequency up to once a week during the previous year. The headaches radiated to the neck and were accompanied by phonophobia and occasionally by nausea, vomiting, and vertigo, therefore fulfilling the criteria for migraine without aura, as proposed by the International Headache Society. Additional examinations (including cardiac diagnostics, duplex sonography of the renal arteries, electroencephalography, screening for thrombophilia and vasculitis) revealed no pathological results.

A duplex sonographic control showed no relevant stenoses of both ICAs. A few days later, the patient developed migrainous headaches, and a long-segment stenosis of the submandibular right ICA was demonstrated on duplex sonography. Oral anticoagulation was started with phenprocoumon. The stenosis progressed to a high-grade stenosis on the next day, but it resolved spontaneously within the following 2 days (Figure 2A and 2B). No intramural hematoma could be detected by MRI.

Regular duplex sonographic controls demonstrated several new episodes of high-grade stenoses alternating weekly in both submandibular ICAs. No clear vessel wall edema could be found, whereas the outer vessel diameter was significantly reduced during stenosis, supporting the presence of vasospasm. During stenosis of the ICA, middle cerebral artery flow velocity (right or left, respectively) was severely reduced. Collateral flow compensation was poor, as indicated by simultaneous activation of primary and secondary collaterals (insufficient crossover flow plus ophthalmic artery). The stenoses were regularly accompanied by migrainous headaches and resolved spontaneously within 1 to 3 days (see Figure 2C for time flow).

Antivasospastic treatment was started with a calcium antagonist (flunarizine, oral dose of 5 mg/d). After a new episode of migrainous headaches and a high-grade stenosis of the left ICA, the flunarizine dose was raised to 10 mg/d, and methylprednisolone (initial dose, 60 mg/d orally, then slowly tapered) and oral magnesium were added to the treatment. No vasospasm occurred during the following 3 weeks. However, ICA vasospasms reoccurred when methylprednisolone was withdrawn, and treatment with oral corticoids was restarted. In subsequent weeks, the frequency of migrainous headaches with concomitant vasospasms of the ICAs was markedly decreased to approximately once a month.

### Discussion

The 2 presented cases demonstrate the diagnostic difficulty when extracranial vasospasm mimics arterial dissection and fibromuscular dysplasia. Vasospasms are a highly dynamic process, and the clinical importance of vasospasm as a cause of stroke might therefore be underestimated. ICA vasospasm has been described in patients with or without a history of migraine. It is not clear whether ICA vasospasms are related to migraine or whether migrainous headache is an epiphenomenon of vasospasm. We propose that recurrent vasospasms of submandibular ICAs with or without migrainous headaches are a distinct entity that can cause ischemic stroke. Some cases might be misdiagnosed as arterial dissection; others might not be detected if examination of the ICAs has not been undertaken during acute vasospasm. Vasospasm of ICAs as a cause of stroke should be considered in patients with recurring ischemic events in the absence of any other explanation, in patients with inconsistent duplex sonographic findings, and when arterial dissection is assumed but no hematoma can be detected by MRI.

Until now, treatment of vasospasms of submandibular ICAs has not been standardized. In case of acute carotid vasospasm, blood pressure should be increased by hypervolemia. Because carotid vasospasms are observed to be recurrent, oral anticoagulation might be used to prevent new ischemic events. Calcium antagonists have been shown to reduce vasospasms after aneurysmal subarachnoid hemorrhage and to be effective in the prophylactic treatment of migraine. However, calcium antagonists...
alone seem to be insufficient in the treatment of vasospasm of the ICA. Patient 1 was treated with calcium antagonists without a beneficial effect. In patient 2, a reduction in the frequency of vasospasms was not achieved by flunarizine alone but only in combination with oral corticoids. Moreover, ICA vasospasms recurred when the treatment with oral corticoids was paused. Similarly, in the patient described by Arning et al., a combined treatment with calcium antagonists and oral corticoids led to a reduction in the frequency of vasospasm. Although the pathomechanism of ICA vasospasm is not yet understood, antiphlogistic treatment seems to be effective.

We hope that in the future more cases of transient vasospasms of ICAs will be correctly diagnosed, which will enhance the probability of finding the correct etiology of this condition as well as optimal treatment for these patients.

Disclosures

None.
References

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