Recurrent Extracranial Carotid Artery Vasospasms
Report of 2 Cases

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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. Extracranial vasospasms are rarely reported and can be provoked by mechanical manipulation or drugs. Spontaneous submandibular vasospasms of the internal carotid arteries (ICAs) have been described in only a few cases, often associated with migraine. Ischemic stroke from reversible arterial vasospasms has been observed in a variety of conditions. 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 showed distal occlusion of the right ICA. Although no intramural hematoma could be detected, dissection of the right ICA was suspected to be the cause of the patient’s stroke.

Two years later, the patient was admitted to the hospital with mild, right-sided hemiparesis and transient vision problems of the left eye. High-grade stenoses of both ICAs were detected on duplex sonography. Two days later, MRA showed only a segmental narrowing of the right ICA but no stenosis of the left ICA. MRA control 4 days later demonstrated severe stenoses of both ICAs (see Figure 1A) but no intramural hematoma. Duplex sonographic examinations during the next few weeks revealed recurring stenoses of both ICAs that resolved spontaneously within hours to days. Depending on the side and severity of the stenoses, especially when both ICA were affected at the same time, intracranial blood flow was markedly reduced, as demonstrated with duplex sonography and perfusion MRI.

The patient was treated with a calcium antagonist (nitrrendipine, oral dose of 20 mg) as an antivasospastic treatment and with phenprocoumon for oral anticoagulation. The duplex sonographic findings continued to change on a daily basis, with regard to both grade and side of stenosis. Conventional digital subtraction angiography with local intra-arterial papaverine injection demonstrated a short-term but distinct widening of the distal ICA stenosis (Figure 1B and 1C). In subsequent weeks, various calcium antagonists were tried, but no reduction of the frequency of ICA vasospasms could be observed.

Patient 2
A 48-year-old woman was admitted to the hospital for lack of motivation, depressive mood, and memory difficulties. MRI revealed a subacute ischemic infarct in the left basal
ganglia. 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 to maintain an activated partial thromboplastin time ratio of 2.0 to 2.5.

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.4–6 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.9 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 hemorrhage10 and to be effective in the prophylactic treatment of migraine.11 However, calcium antagonists

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**Figure 1.** A, MRA demonstrates high-grade stenoses of both ICAs. B and C, Digital subtraction angiography of the right ICA before (B) and after (C) papaverine injection which leads to an increase of the vessel diameter.
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., 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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