

PARATRIGEMINAL NEURALGIA THAT IMPROVED WITH TREATMENT OF HYPERTENSION IN A PATIENT WITH RAEDER'S SYNDROME AFTER CAROTID ARTERY STENTING

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Background: Although there have been reports of Raeder's syndrome developing after carotid artery dissection, to our knowledge, no case of Raeder's syndrome occurring after carotid stenting has been reported.

Case Report: A 46-year-old man was urgently treated with a self-expanding stent for idiopathic right carotid artery dissection. However, the patient complained of moderate oppressive pain in the right orbit and forehead immediately after stent placement. Further examination revealed a right eye miosis and right ptosis. No anhidrosis was noted. A diagnosis of Raeder's syndrome was made based on partial Horner's syndrome and pain in the first branch of the trigeminal nerve. We noticed that he had hypertension and started antihypertensive medication. After starting the antihypertensive medication, his blood pressure stabilized, and his periorbital pain disappeared dramatically as his blood pressure fell. However, Horner's symptoms (miosis and ipsilateral ptosis) were still present.

Conclusion: We experienced a case of Raeder's syndrome that occurred after stenting of the carotid artery. Neuralgia improved by treatment of hypertension. It should be noted that the paratrigeminal neuralgia of Raeder's syndrome after carotid stenting can be caused by hypertension.

Key words: Antihypertensive medication, Horner's symptoms, Raeder's syndrome, stenting of the carotid artery

BACKGROUND

Raeder's syndrome is a variant of Horner's syndrome with trigeminal nerve dysfunction and cluster headaches. It is usually thought to be caused by inflammation or mass lesions of sympathetic fibers of the petrous or cavernous carotid artery and nearby trigeminal fibers traversing the floor of the middle fossa. Raeder's syndrome has been reported to occur after carotid artery dissection (1,2). We present a case of paratrigeminal

neuralgia that improved with treatment of hypertension in a patient with Raeder's syndrome that developed after stent-supported percutaneous angioplasty for carotid artery dissection.

CASE

A 46-year-old man was referred to another facility for a sudden onset of quadrant hemianopsia. He was in good health up to that point and had no medical history

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or history of trauma. Angiography performed for diagnosis showed severe stenosis of the right internal carotid artery with peel and string sign (Fig. 1). Collateral blood flow from the left carotid artery was good. Clinically, the patient presented with acute intermittent quadrant hemianopsia of the right eye, which was thought to be caused by partial occlusion of the central retinal artery. Idiopathic right carotid artery dissection was diagnosed and urgently treated with a self-expanding stent. Three overlapping stents were required to obtain an angiographically smooth contour in the true lumen (Fig. 2). Doppler ultrasonography of the carotid artery after stenting showed no stenosis, dissection, intramural hematoma, or stent migration. However, the patient complained of moderate oppressive pain in the right orbit and forehead immediately after stent placement. His corneal and facial sensation was normal. There were no other sequelae and the patient was discharged on the fifth postoperative day.

After discharge, the patient's periorbital pain worsened and the patient was referred to a nearby ophthalmologist. Further examination at the hospital revealed a right eye miosis and right ptosis. No anhidrosis was noted. A diagnosis of Raeder's syndrome was made based on partial Horner's syndrome and pain in the first branch of the trigeminal nerve. Reexamination by

magnetic resonance imaging of the head showed no obvious abnormalities such as occupational lesions in the skull or orbit, infarct lesions, or vascular stenosis.

Two months after stent placement, the ophthalmologist referred the patient to our pain clinic for treatment of ipsilateral periorbital pain. The patient's pain was characterized by a continuous, squeezed, oppressive, and nonpulsatile pain only on the right side of the forehead and around the eye.

There was also severe pain in the same area about once a week. It was not accompanied by eye watering, nasal congestion, or swelling around the eyes. He had a history of postherpetic neuralgia of the first branch of the right trigeminal nerve, but the symptoms of neuralgia had resolved long ago. He had no other medical history and was not taking any medications. He was started on tramadol hydrochloride and acetaminophen combination tablets.

After starting the medication, his pain was reduced to about half of its subjective level. However, the pain persisted and interfered with his daily activities. While continuing the medication, we noticed that his blood pressure was above 180 mmHg. He had never been diagnosed with hypertension before. Further examination revealed that his systolic blood pressure was above 160 mmHg on waking and at bedtime. A diagnosis of

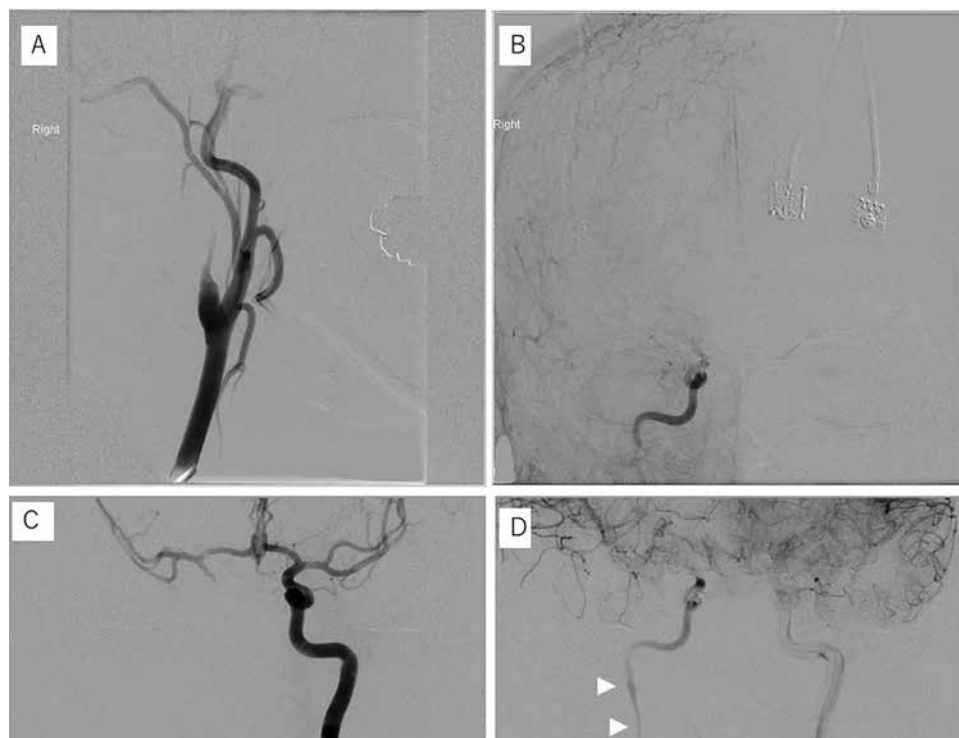
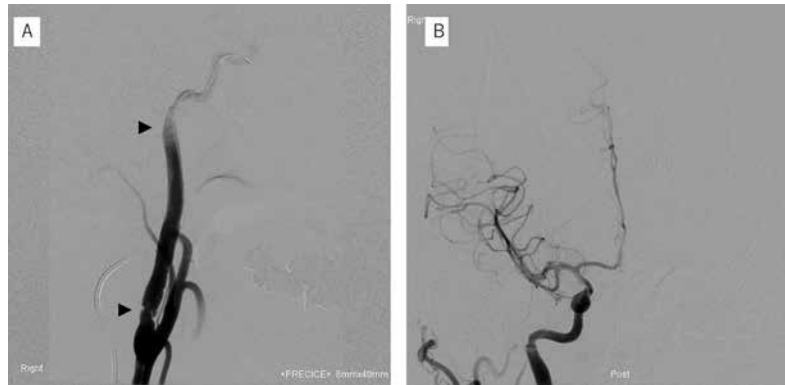


Fig. 1. A, B) Right common carotid artery angiograms show severe stenosis of the proximal ICA due to dissection and poor visualization of the intracranial arteries. C, D) Left common carotid artery angiograms with proximal balloon test occlusion of the right common carotid artery show good collateral flow of the right hemisphere from the left ICA and long dissected segment of the right cervical ICA in capillary phase (white arrow heads, D).

Fig. 2. A, B) Angiograms just after self-expanding stents placement show restoration of right ICA and clear visualization of intracranial arteries. Three overlapped stents are placed from the proximal cervical ICA to the proximal petrous ICA (black arrow heads, A).

Abbreviations: ICA, internal carotid artery



hypertension was made and antihypertensive medication was started. After starting the antihypertensive medication, his blood pressure stabilized to 120 mmHg and his periorbital pain disappeared dramatically as his blood pressure fell. After the pain disappeared, the dose of analgesia was gradually reduced, making sure that there was no recurrence of pain. During this time, his blood pressure remained stable on oral antihypertensive medication. After approximately one month, the tramadol and acetaminophen combination tablets were discontinued, but the pain did not recur. The antihypertensive medication was continued and the patient was referred to another hospital with no pain, and treatment at our hospital for 4 months after stenting was terminated. At this point, Horner's symptoms (miosis and ipsilateral ptosis) were still present.

DISCUSSION

Raeder's syndrome was first described by a Norwegian neurologist in 1924. This condition is also known as paratrigeminal syndrome. Clinically, it is characterized as unilateral migraine or cluster-like headache, with pain distributed to the first (V1) and second (V2) segments of the ipsilateral trigeminal nerve and accompanied by incomplete Horner's syndrome (miosis, ptosis, and preserved facial sweating). Raeder's syndrome is usually attributed to inflammation or mass lesions disrupting the sympathetic fibers of the petrous or cavernous carotid artery and nearby trigeminal fibers traversing the floor of the middle fossa. Although there have been reports of Raeder's syndrome developing after carotid artery dissection (1,2), to our knowledge, no case of Raeder's syndrome occurring after carotid stenting has been reported. In the present case, we experienced a case of Raeder's syndrome that developed after carotid

stenting and the paratrigeminal neuralgia resolved with treatment of hypertension.

The patient complained of mild periorbital and forehead oppressive pain from the day of stenting. Fay et al (3) reported that the stimulation of the internal carotid artery near the bifurcation causes an ipsilateral craniofacial pain of cluster headache type. In addition, Milena et al (4) performed a pharmacological pupillary examination in 37 patients who underwent internal carotid endarterectomy and showed that surgical damage to the sympathetic plexus was associated with postoperative cluster-like headache. Our patient's headache was located in the periorbital region, as seen in cluster headache, but the headache was continuous and was not accompanied by eye redness, nasal congestion, or swelling around the eyes. After his blood pressure normalized, the periorbital pain disappeared. This suggests that his pain was associated with hypertension. However, according to the International Classification of Headache Disorders, headaches attributable to arterial hypertension are often characterized as bilateral and pulsatile. The patient's pain in the present case could have been characterized by both cluster headache and hypertension-related headache. Increased blood flow to the orbit due to hypertension and dilation of the sinusoidal carotid artery may have caused unilateral periorbital pain. The pain disappeared as blood pressure normalized and stimulation of the sympathetic plexus decreased.

On the other hand, Horner's syndrome remained after the blood pressure became normal and the pain disappeared. Horner's syndrome has been reported to occur secondary to carotid artery pseudoaneurysms (5), true aneurysms (6), atherosclerosis (7), subintimal hematoma within atherosclerotic plaques (8), and carotid artery

dissections (9). In patients with carotid artery dissection, 40% to 50% of them will develop ipsilateral Horner syndrome (10,11). There has been one case report (12) and one retrospective study of the development of Horner's syndrome after carotid stenting (13), with the possible causes being stent dilation and compression by a hematoma. Ringer et al (12) mentioned that the stretching of the artery wall after stenting may result in stretching the sympathetic fibers surrounding the internal carotid artery. In this report, the stent used was a self-expanding stent and was oversized relative to the native lumen diameter. However, in our case, the stent was not oversized on the postoperative computed tomography scans. Furthermore, Rosenkranz et al (13) showed that Horner's syndrome was significantly associated with the appearance of ipsilateral carotid artery wall hematoma after the intervention. In our

case, ultrasonography of the carotid artery after stent placement revealed no evidence of carotid artery wall hematoma. For these reasons, we suspect that in the present case, the sympathetic fibers were impaired from the initial dissection injury, and that the additional stretch caused by stent placement and dilation may have induced Horner's syndrome. Treatment of hypertension improved paratrigeminal neuralgia but not Horner's syndrome.

CONCLUSION

In the present case, we experienced a case of Raeder's syndrome that occurred after stenting of the carotid artery. Neuralgia improved by treatment of hypertension. It should be noted that the paratrigeminal neuralgia of Raeder's syndrome after carotid stenting can be caused by hypertension.

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