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Carotidynia presenting with acute ischemic stroke after carotid sinus massage

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Carotidynia is characterized by unilateral neck pain around the carotid artery. We describe a 50-year-old woman who presented with transient left-side weakness and right-side neck pain. She frequently massaged the uncomfortable neck area during the symptomatic course of the condition. Magnetic resonance imaging revealed multifocal cerebral infarctions and a carotid intramural thrombus ipsilateral to the carotidynia.Long-term carotidynia might result in the involvement of an intramural thrombus and intimal disruption, and ischemic stroke after carotidynia may be provoked by carotid sinus massage.

Key words: Carotid artery; Pain; Massage

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Carotidynia is characterized by unilateral tenderness around the carotid artery, especially near the carotid bifurcation, as first described in 1927 by Temple Fay.[1] The pain often radiates to the ipsilateral mandible, cheek, eye, and ear, and it may occur continuously or periodically. Inflammatory components demonstrated in histological examinations have been implicated in carotidynia, but the findings have not been definitive.[2] There are some case reports of fairly consistent magnetic resonance imaging (MRI) and computed tomography findings suggestive of carotid inflammation around the carotid bifurcation.[3-6] In rare cases carotidynia may present with a carotid arterial thrombus or disruption of the intimal plaque.[5,6] Furthermore, there is little information available about carotidynia accompanied with acute ischemic stroke.

Here we present a case of carotidynia with acute ischemic stroke that was related to previous carotid sinus massage (CSM) performed by the patient.

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CASE

A 50-year-old woman was admitted to the emergency room with transient left-side weakness and mild dysarthria with a duration of 90 min. She had accompanying pain in the right mandibular area that was radiating to the ipsilateral neck and had been present for more than 3 weeks. The pain was not accompanied by focal swelling or redness, but the uncomfortable symptoms prompted the patient to massage the painful area frequently herself. She had stable vital signs (blood pressures of 116/59 mmHg and heart rate of 69 beats/min) except for a mild fever of 37.7℃. Blood laboratory examinations revealed no specific findings except for an elevated erythrocyte sedimentation rate of 44 mm/h.

Her initial neurologic symptoms recovered on the admission day (with the exception of neck pain), which led to a score on the National Institutes of Health Stroke Scale of 0. Diffusion-weighted imaging revealed multifocal infarctions in the right basal ganglia, frontal cortex, and parietal cortex (Fig. 1A). Volume-rendering imaging showed a filling defect in the long segment of the common carotid artery (CCA) and the internal carotid artery (ICA) (Fig. 1B). An axial source image obtained in time-of-flight carotid magnetic resonance angiography (MRA) revealed a massive intraluminal filling defect in both the right CCA and ICA (Fig. 1C). She was initially treated with dual antiplatelet agents and anti-inflammatory drugs.

On the fourth admission day her neck pain was relieved,

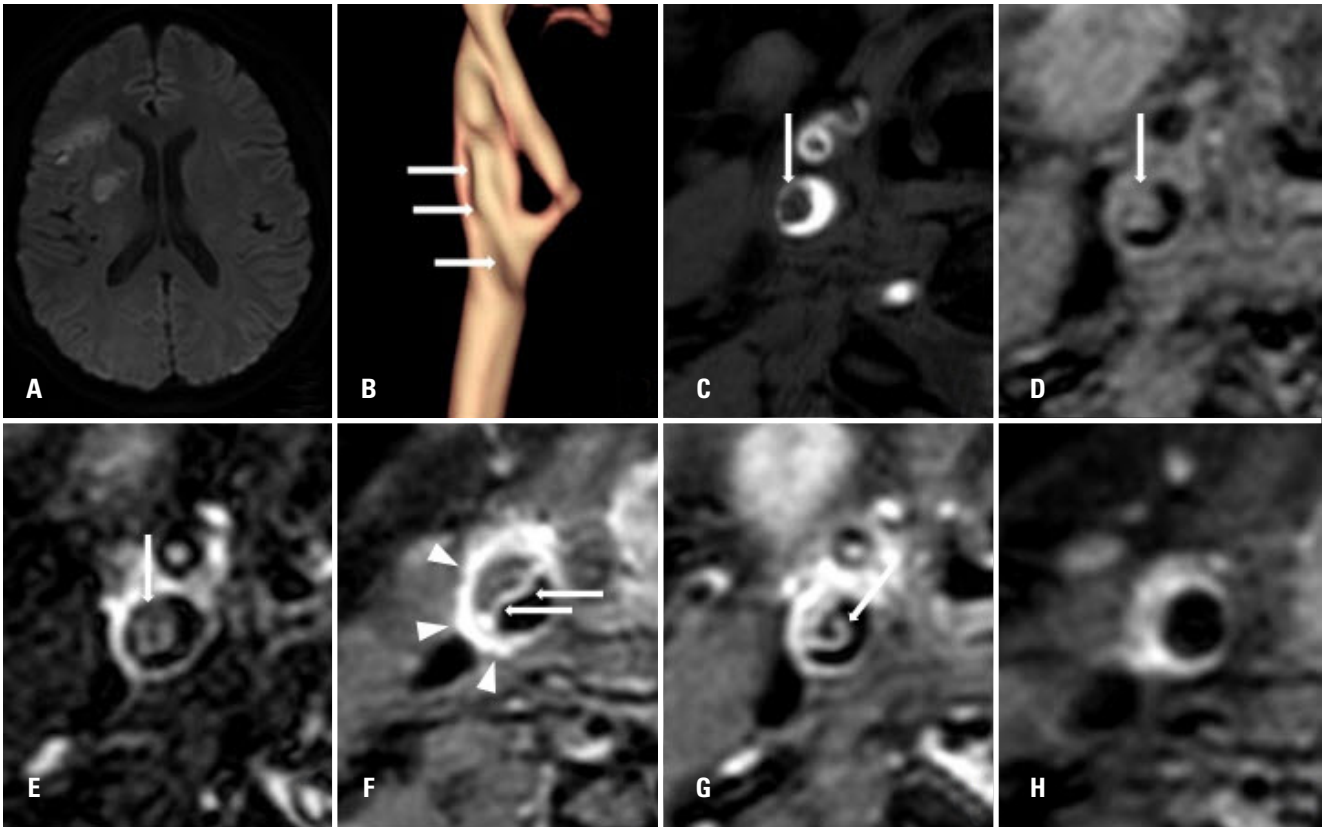

Fig. 1. A 50-year-old woman with a transient left-side weakness and mild dysarthria. (A) Diffusion-weighted imaging showing multifocal infarctions in the right basal ganglia, frontal cortex, and parietal cortex. (B) Volume-rendering imaging showing a filling defect in the long segment of the CCA and ICA (arrows). (C) Axial source image of time-of-flight carotid MRA showing a massive intraluminal filling defect in the right CCA (arrow). (D) Axial image of nonenhanced T1-weighted carotid MRI showing the intravascular thrombus without an intramural plaque or intimal flap (arrow). (E) Axial image of nonenhanced T2-weighted carotid MRI showing the intravascular thrombus without an intramural plaque or intimal flap (arrow). (F) Contrast-enhanced MRI of the right CCA showing the circumferential enhancement (arrowheads) with a mural wall thrombus (arrows). (G) Contrast-enhanced MRI of the right ICA showing the intimal disruption (arrow). (H) Contrast-enhanced axial imaging performed 10 days after the initial carotid plaque MRI showing complete resolution of the mural wall thrombus and intimal disruption and a marked decrease in the circumferential carotid wall enhancement. CCA, common carotid artery; ICA, internal carotid artery; MRA, magnetic resonance angiography; MRI, magnetic resonance imaging.

and the patient had no specific clinical symptoms. She was evaluated using high-resolution carotid plaque MRI. Axial images of non-enhanced T1 and T2-weighted MRI revealed an intravascular thrombus without an intramural plaque or intimal flap (Fig. 1D, E). Vascular inflammation was suspected from contrast-enhanced MRI showing circumferential enhancement on the right distal CCA and carotid bifurcation. Other specific findings were a mural wall thrombus (Fig. 1F) and intimal disruption on the inside of the circumferential enhancement area (Fig. 1G), which was also confirmed by ultrasonography of the carotid artery. She was treated with prednisolone (20 mg) once daily, anti-inflammatory drugs, and continuous heparinization.

Follow-up contrast MRI performed 10 days after the initial imaging study, showed complete resolution of the mural wall thrombus with intimal disruption and a marked decrease of the circumferential carotid wall enhancement (Fig. 1H). She was discharged with no specific neurologic or clinical symptoms on day 20 after admission.

DISCUSSION

Dissection of the carotid artery might show an intimal flap and double lumen, but carotid artery dissection accompanied by inflammation of vessel wall and its surrounding tissues has not been well characterized. In the present case, the crescent-shape of the irregular enhancement on the carotid wall and perivascular tissues was suggestive of carotidynia. Neuroimaging findings of a double lumen in the carotid artery might have been due to secondary dissection associated with inflammatory changes. We diagnosed the present case as carotidynia based on the criteria of the International Headache Society (IHS).[1] In addition, the patient had three other specific findings that are unusual among the well-known characteristics of carotidynia.

Firstly, her ischemic stroke occurred with carotidynia. The general consensus of imaging findings of carotidynia is 1) continuous involvement of the distal CCA, bifurcation, or proximal ICA, 2) concentric enhancement of the carotid wall and surrounding tissue, and 3) slight luminal narrowing.[3-7] The MRA findings of our patient were consistent with the usual findings but also showed a mural thrombus and intimal disruptions. A few cases of carotidynia with thrombus formation have been reported, but none of them had the complication of ischemic stroke.[4,5] Dissection of the carotid artery might also show the intimal flap and double lumen. However, it has not been well described that carotid artery dissection causes inflammation of the vessel wall and surrounding tissues.

The second unusual finding in the present case is the duration of pain and thrombus formation. The patient had suffered from 3 weeks of neck pain, which was longer than the IHS criteria. Previous reports suggest that severe carotid inflammation is sufficient for thrombus formation due to the possibility of perivascular inflammation propagating to the lumen.[4,5] The longer duration of carotidynia in the present case might have aggravated perivascular inflammation with the mural thrombus, which could have contributed to ischemic stroke.

Lastly, we propose that CSM is another important triggering factor of ischemic stroke. Due to the possible eventual complication of CSM, there are well-known contraindications: known carotid stenosis, transient ischemic attack, history of stroke within the previous 3 months, and audible bruit over the carotid arteries.[8] Moreover, Currò et al.[9] reported a woman who was diagnosed with ischemic stroke in the right middle cerebral artery after CSM without contraindication for CSM. We propose that the CSM was another important triggering factor of ischemic stroke in that patient.

One limitation of the present study to consider is that the reported case is not entirely consistent with the IHS criteria for carotidynia—the IHS suggests the absence of structural abnormality for a diagnosis of carotidynia as an idiopathic etiology, which differs from the present case.

In conclusion, long-term carotidynia may present with an intramural thrombus and intimal disruptions. Moreover, applying CSM on the symptomatic side of carotidynia could increase the probability of ischemic stroke.

REFERENCES

1. Spiller WG. Philandelphia Neurological Society: regular meeting, Jan. 28, 1927. Arch Neurol Psychiatry 1927;18:308.
2. Upton PD, Smith JG, Charnock DR. Histologic confirmation of carotidynia. Otolaryngol Head Neck Surg 2003;129:443-444.
3. Schaumberg J, Eckert B, Michels P. Carotidynia : magnetic reso-

nance imaging and ultrasonographic imaging of a self-limiting disease. Clin Neuroradiol 2011;21:91-94.
4. Burton BS, Syms MJ, Petermann GW, Burgess LP. MR imaging of patients with carotidynia. AJNR Am J Neuroradiol 2000;21:766-769.
5. Matsubara H, Takeshita K, Nakamura S, Murohara T, Miyachi S. Carotidynia with carotid arterial thrombosis. Ann Intern Med 2012;157:917-919.
6. Woo JK, Jhamb A, Heran MK, Hurley M, Graeb D. Resolution of existing intimal plaque in a patient with carotidynia. AJNR Am J Neuroradiol 2008;29:732-733.
7. Kuhn J, Harzheim A, Horz R, Bewermeyer H. MRI and ultrasonographic imaging of a patient with carotidynia. Cephalalgia 2006;26:483-485.
8. Paling D, Vilches-Moraga A, Akram Q, Atkinson O, Staniland J, Paredes-Galán E. Carotid sinus syndrome is common in very elderly patients undergoing tilt table testing and carotid sinus massage because of syncope or unexplained falls. Aging Clin Exp Res 2011;23:304-308.
9. Currò DR, Roscia G, Turri E, Dall'Ora E, Sansone S, Stockner I, et al. Acute ischemic stroke complicating carotid sinus massage in the absence of carotid artery disease and failure of thrombolytic therapy. Minerva Med 2010;101:193.