

[CASE REPORT]

Vertebral Artery Stump Syndrome

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Abstract:

Carotid stump syndrome is a well-documented embolic source for ischemic stroke. However, few cases have been reported of a similar condition - termed vertebral artery stump syndrome - which affects the posterior circulation after vertebral artery origin occlusion. We herein report a case of infarction of the right superior cerebellar artery and left posterior inferior cerebellar artery territories due to vertebral artery stump syndrome. In this interesting case, a turbulent flow at the distal side of the vertebral artery occlusion was captured on ultrasonography, and was identified as the probable mechanism of vertebral artery stump syndrome.

Key words: vertebral artery stump syndrome, ultrasonography, spiral turbulent flow, embolic ischemic stroke, bilateral cerebellar infarction

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Introduction

Carotid stump syndrome is a well-documented embolic source for ischemic stroke (1-4). However, few reports have so far described a parallel syndrome, which causes posterior circulation ischemic stroke after occlusion of the origin of the vertebral artery (VA). This type of ischemic stroke results from the occlusion of the VA and has been termed "vertebral artery stump syndrome" (2-4). To the best of our knowledge, the possible mechanisms underlying vertebral artery stump syndrome have not previously been captured on imaging.

In this report, we describe how we clearly identified the cause of the embolism in a patient with vertebral artery stump syndrome using ultrasonography.

Case Report

A 66-year-old man presented with dysarthria. He had suffered a cerebral infarction 16 years previously, and remained paralyzed on his left side. He became aware of dysarthria in the morning and although he experienced no new paralysis, he was admitted to the neurology department of our hospital 6 hours later because of sustained dysarthria. The patient

had a history of hypertension, diabetes, and dyslipidemia, and was receiving treatment with oral hypoglycemic agents. He reported habitually smoking 40 cigarettes per day for 30 years.

On admission (day 1), a physical examination revealed hypertension (213/93 mmHg) and a normal pulse rate and rhythm. He was alert and exhibited cerebellar speech and left hemiparesis. However, the left hemiparesis appeared to be related to the previous cerebral infarction. We were unable to examine him for cerebellar symptoms because of his left hemiparesis. On the left side, the tendon reflexes were exaggerated and a pathological plantar reflex was elicited. Diffusion-weighted imaging (DWI) on admission showed an acute ischemic stroke in the right cerebellar hemisphere of the superior cerebellar artery (SCA) territory and the left cerebellar hemisphere of the posterior inferior cerebellar artery (PICA) territory (Fig. 1a, b). The basilar artery and both VAs were well visualized on magnetic resonance (MR) angiography. Anti-platelet therapy with clopidogrel (75 mg/day), together with an intravenous infusion of edaravone, ozagrel, low-molecular dextran, and statin [low density lipoprotein cholesterol (LDL-C) on admission: 169 mg/dL] was administered to treat the acute cerebral infarction.

On day 6, three-dimensional computed tomographic (CT) angiography revealed an occlusion at the origin of the left

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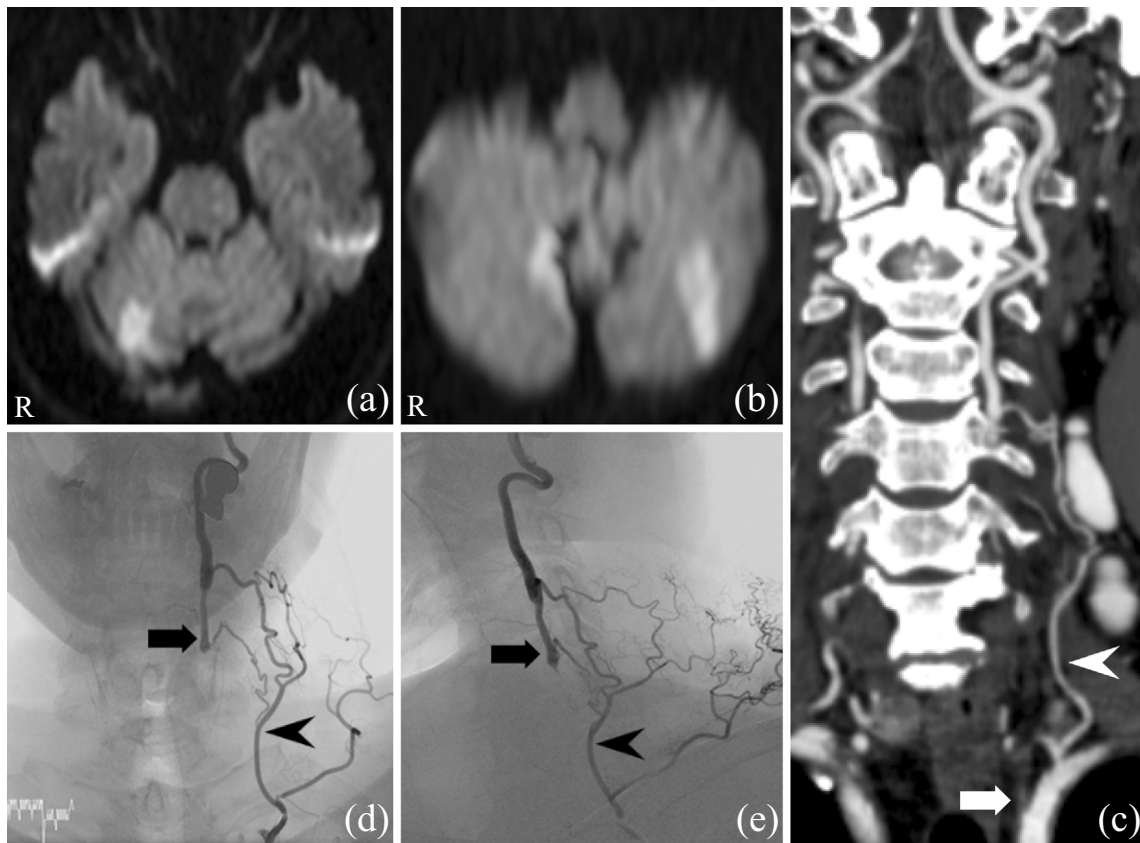


Figure 1. The imaging findings. (a, b) Diffusion-weighted imaging on admission (day 1) showed hyperintense lesions in the right SCA territory and the left PICA territory. (c) Three-dimensional CT angiography showed occlusion at the origin of the left VA (white arrow) with distal inflow via the deep cervical artery at the C4 and C5 level (white arrowhead). The left deep cervical artery angiogram (d: AP view, e: lateral view) showed a turbulent flow (arrow) at the proximal end of the left patent VA via the collateral flow at the C5 level and the distal antegrade collateral flow of the left patent VA via the deep cervical artery (arrowhead) at the C4 and C5 level.

VA with distal inflow via the deep cervical artery at the C4 and C5 level (Fig. 1c). On day 12, we performed cerebral angiography. The left VA (injected via the left subclavian artery) demonstrated the occlusion of the VA at its origin and revealed collateral circulation. At the C5 level, the left deep cervical artery angiogram showed both a turbulent flow at the proximal end of the left patent VA (via the collateral flow from the deep cervical artery), and distal antegrade flow of the left patent VA (via the collateral flow from the deep cervical artery) (Fig. 1d, e). Ultrasonography (Ultrasonography System; ProSound ALPHA7, Linear probe 7.5 MHz, Hitachi, Tokyo, Japan) also showed occlusion at the origin of the left VA (Fig. 2a), and revealed collateral flow from the deep cervical artery (Fig. 2b). Spiral turbulent flow (Fig. 2c) and antegrade flow (Fig. 2d) were observed on the distal side of the site of the left VA occlusion.

The plasma brain natriuretic peptide (BNP) level was normal, and transthoracic echocardiography and 24-h electrocardiography (ECG) monitoring showed no abnormalities indicating the existence of cardiac embolic sources. We therefore considered the source of the embolism to be vertebral artery stump syndrome due to occlusion of the VA. After completing intravenous infusion and rehabilitation therapy,

the patient's condition improved. No recurrent ischemia was observed and the patient's scanning speech resolved. On day 24, he was discharged from hospital and returned home. The clopidogrel treatment was continued. A follow-up examination at 11 months revealed that no further events had occurred.

Discussion

Among the patients with posterior circulation cerebral infarcts after acute stroke, 48.7% of the patients have a cerebellar infarct; 64.3% of these patients have unilateral lesions, while 35.7% have bilateral lesions (5). Thus, bilateral cerebellar infarcts account for 17.4% of all posterior circulation ischemic strokes. Bilateral cerebellar infarcts are therefore considered to be relatively common. According to Wang et al., bilateral cerebellar infarcts frequently involve the PICA and SCA territories; our patient had a cerebellar infarct in each of these territories. Embolism, resulting from large artery atherosclerosis, is an important stroke mechanism in such cases (5).

We ruled out a cardiogenic embolism in our patient based on his normal plasma BNP level and the absence of any ab-

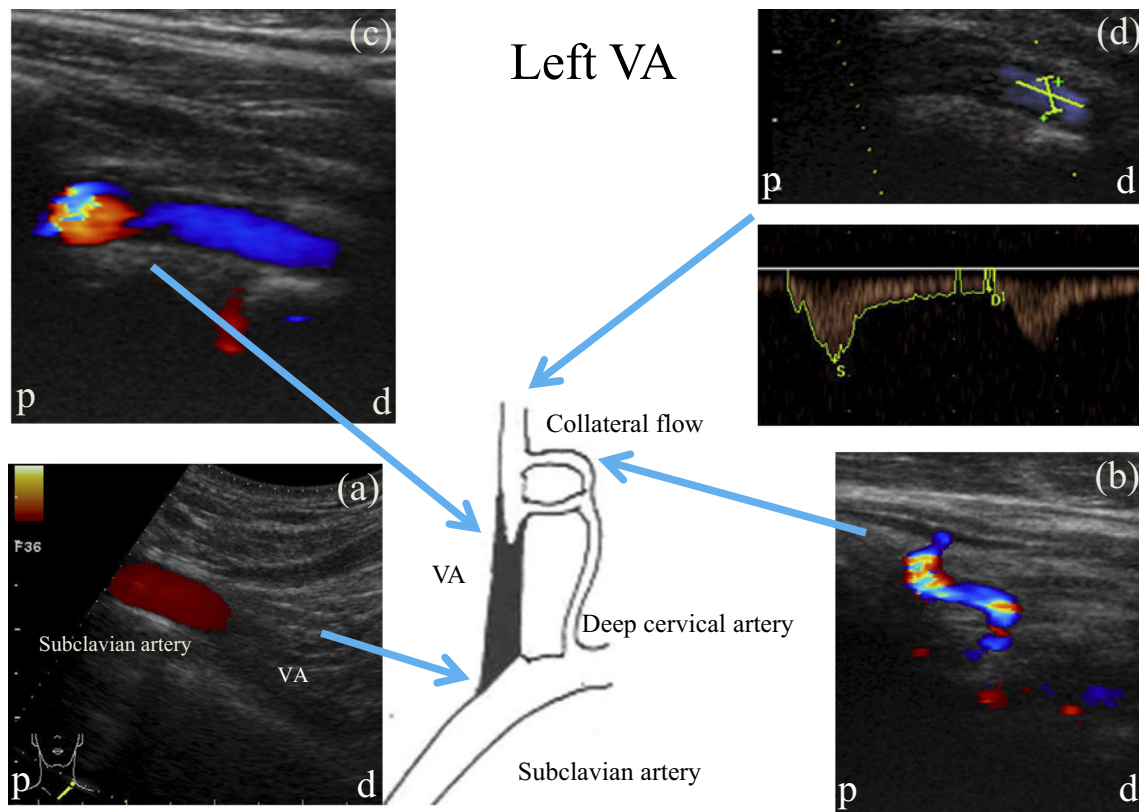


Figure 2. The ultrasonography findings. Ultrasonography showed occlusion at the origin of the left VA (a) and revealed collateral flow from the deep cervical artery (b). Spiral turbulent flow (c) and antegrade flow (d) on the distal side of the site of left VA occlusion. p: proximal side, d: distal side

normalities on transthoracic echocardiography or 24-h ECG monitoring. The imaging studies that we performed included carotid ultrasonography, three-dimensional CT angiography and cerebral angiography; the results of these examinations revealed atherosclerotic occlusion of the left VA at its origin. Accordingly, we diagnosed vertebral artery stump syndrome due to left VA occlusion.

Carotid stump syndrome is defined as the persistence of cerebral or retinal ischemic symptoms after the occlusion of an ipsilateral internal carotid artery, and it is well known to be a potential source of micro-emboli (1). Further ischemic episodes are not expected after the occlusion of the arterial flow due to the lack of a flow conduit to carry the embolus. However, in patients with carotid stump syndrome, ongoing ischemic events may continue, despite the artery being occluded, because the collateral flow via the external carotid artery branches can carry proximal or distal emboli to the intracranial internal carotid artery (ICA) (2). Kato et al. called this condition proximal stump occlusion. In contrast, they defined distal stump as the occlusion of the common carotid artery, where ischemic episodes continued to occur via the patent ICA (6). Turbulent flow in both stumps can cause platelet-fibrin aggregation and may theoretically lead to embolization (6). Although carotid stump syndrome is a well documented embolic source for ischemic stroke, there have been few reports on vertebral artery stump syndrome, which is a similar syndrome in the posterior circulation that

occurs after occlusion at the origin of the VA (2). The diagnostic criteria for vertebral artery stump syndrome are as follows: (a) acute ischemic stroke in the posterior circulation; (b) occlusion at the origin of the VA identified on MR angiography, duplex ultrasonography, CT angiography, and/or conventional angiography; (c) the presence of distal antegrade flow in the ipsilateral VA; and (d) the absence of other causes of ischemic stroke (3).

It has been suggested that a thrombus produced immediately after occlusion at the origin of the VA might cause ischemic stroke (3). The following mechanism may explain the pathogenesis of ischemic stroke in patients with vertebral artery stump syndrome: (a) vertebral artery stump syndrome after ipsilateral occlusion of the origin of the VA can be caused by the distal limit of the propagated thrombus; and (b) the occurrence of ischemic events after the occlusion of the VA may be associated with emboli from the stagnating clot fragment and a low-flow state due to the collateral circulation via the deep cervical arteries, especially in the absence of any other embolic sources or hematological disorders (3). To date, 10 cases of vertebral artery stump syndrome involving the collateral pathway have been reported (2, 3, 7). In 9 of these, the collateral tract was derived from the deep cervical artery; in 1 case, it was derived from the ascending cervical artery. To the best of our knowledge, in cases where the collateral pathway was derived from the deep cervical artery, the inflow to the VA came via

1-4 blood vessels, and the sites of inflow were heterogeneous (C1 to C6). Our patient had a collateral pathway to the vertebral artery at the C4 and C5 level. We angiographically diagnosed the origin of the collateral route as the deep cervical artery.

However, few reports have clarified the exact cause of ischemic stroke in vertebral artery stump syndrome. Our case is valuable in that it shows turbulent flow on the distal side of the occlusion of the VA and this turbulence appears to be the cause of the embolism in vertebral artery stump syndrome.

It is not yet known whether the best treatment for this condition is antiplatelet therapy or anticoagulation therapy. Kawano et al. reported that the rate of relapse in patients receiving antiplatelet therapy was high in comparison to those receiving anticoagulation therapy, and they recommended anticoagulation therapy (3, 4). However, in the present case, we selected antiplatelet therapy because we considered that an artery-to-artery embolism (due to atherosclerotic changes) was responsible for the ischemic event, based on the medical history of the patient. No further events occurred during the 11-month follow-up period. A few reports have described the application of endovascular treatment in cases in which medical treatment with antiplatelet therapy or anticoagulation therapy was unsuccessful. For example, Nguyen et al. (2) reported that coil embolization was performed to exclude the embolic source. Nii et al. (7) reported that the occluded VA was penetrated by a guidewire and subsequently revascularized by endovascular angioplasty using a stent. Further studies are required to determine whether antiplatelet therapy, anticoagulation therapy, endovascular therapy, or a combination of these offer the best treatment for patients with posterior circulation ischemic stroke resulting from vertebral artery stump syndrome.

Conclusion

When VA occlusion is detected in patients with posterior circulation ischemic stroke, physicians should consider the possibility of vertebral artery stump syndrome. This valuable case revealed the presence of turbulent flow on ultrasonography. This appears to be the mechanism underlying vertebral artery stump syndrome. It is important to select an appropriate therapy according to the pathogenesis of ischemic stroke.

The authors state that they have no Conflict of Interest (COI).

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