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A Case Report of Locked-in Syndrome Due to Bilateral Vertebral Artery Dissection After Cervical Spine Manipulation Treated by Arterial Embolectomy

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Abstract: Cervical spine manipulation (CSM) is a commonly spinal manipulative therapies for the relief of cervical spine-related conditions worldwide, but its use remains controversial. CSM may carry the potential for serious neurovascular complications, primarily due to vertebral artery dissection (VAD) and subsequent vertebrobasilar stroke. Here, we reported a rare case of locked-in syndrome (LIS) due to bilateral VAD after CSM treated by arterial embolectomy.

A 36-year-old right-handed man was admitted to our hospital with numbness and weakness of limbs after treating with CSM for neck for half an hour. Gradually, although the patient remained conscious, he could not speak but could communicate with the surrounding by blinking or moving his eyes, and turned to complete quadriplegia, complete facial and bulbar palsy, dyspnea at 4 hours after admission. He was diagnosed with LIS. Then, the patient was received cervical and brain computed tomography angiography that showed bilateral VAD. Aortocranial digital subtraction angiography showed vertebrobasilar thrombosis, blocking left vertebral artery, and stenosis of right vertebral artery. The patient was treated by using emergency arterial embolectomy and followed by antiplatelet therapy and supportive therapy in the intensive care unit and a general ward. Twenty-seven days later, the patient's physical function gradually improved and discharged but still left neurological deficit with muscle strength grade 3/5 and hyperreflexia of limbs.

Our findings suggested that CSM might have potential severe side-effect like LIS due to bilateral VAD, and arterial embolectomy is an important treatment choice. The practitioner must be aware of this complication and should give the patients informed consent to CSM, although not all stroke cases temporally related to SCM have pre-existing craniocervical artery dissection.

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Abbreviations: CSM = cervical spine manipulation, CT = computed tomography, CTA = CT angiography, DSA = digital subtraction angiography, LIS = locked-in syndrome, MRI = magnetic resonance imaging, VAD = vertebral artery dissection.

INTRODUCTION

Spinal manipulative therapy was defined by World Health Organization as “includes all procedures where the hands or mechanical devices are used to mobilize, adjust, manipulate, apply traction, massage, stimulate, or otherwise influence the spine and paraspinal tissues with the aim of influencing the patient's health.”¹ Cervical spine manipulation (CSM) is one popular type of spinal manipulative therapies for the relief of cervical spine-related conditions worldwide, which is considered a high-level skill of manual therapy that demands bimanual coordinated rhythmical movements.² Currently, CSM has emerged as one of the leading forms of alternative and complementary treatment for neck pain from cervical spinal disease.³ However, serious complications of CSM continue to be reported.⁴ CSM may carry the potential for serious neurovascular complications, primarily due to vertebral artery dissection (VAD) and subsequent vertebrobasilar stroke.⁵ Here, we reported a rare case of locked-in syndrome (LIS) due to bilateral VAD after CSM treated by arterial embolectomy.

CONSENT

Written informed consent was obtained from the patient before and after all procedures.

CASE PRESENTATION

A 36-year-old right-handed man presented with numbness and weakness of limbs was transferred to emergency department in our hospital. He had been in good health before. One day before admission, the patient suffered from mild neck pain with no focal neurological deficits and 2 hours before he visited a chiropractor and underwent CSM in a private clinic. However, during the period of CSM, the patient's pain deteriorated suddenly half an hour before admission. At that time, the patients complained numbness and hemiplegia in the right side of limbs. Then the patient was sent to the emergency room of our hospital immediately.

The patient underwent a number of investigations. The 4 main vital signs routinely monitored as follows: temperature was 37°C, heart rate was 80 beats per minute, blood pressure was 132/88 mmHg, and respiratory rate was 26 breaths per minute. Respiratory, cardiac, and abdominal examinations were

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within normal limits. Routine blood screens showed white blood cell count was $13.1 \times 10^9 L^{-1}$ and other items were unremarkable. Coagulation times, renal and liver function tests, and electrolytes were within normal limits. The cervical vertebra computed tomography (CT) showed mild intervertebral disc hernia of C3/C4, C4/C5, and brain CT scan was normal. Gradually, although the patient remained conscious, he could not speak but could communicate with the surrounding by blinking or moving his eyes, and turned to complete quadriplegia, complete facial and bulbar palsy, dyspnea at 4 hours after admission. The patient developed LIS. The repeated brain CT scan showed the hyperdense basilar artery sign and cervical vertebra CT scan showed no evident abnormality in cervical cord. (Figure 1A, B). Owing to the symptoms persisting and progressing, brain CT angiography (CTA) examination was performed at 8 hours after admission. The cervical and brain CTA showed (Figure 2A): bilateral VAD. The patient was transferred to the intensive care unit to prepare for the emergency embolectomy with intubated but without mechanical ventilation.

Aortocranial digital subtraction angiography (DSA) under general anesthesia was performed immediately and endovascular treatment was taken as well. A 6-Fr introducer sheath was placed into the right femoral artery via a percutaneous route. Aortocranial DSA shows vertebralbasilar thrombosis, blocking left vertebral artery, and stenosis of right vertebral artery (Figure 2B, C). After emergency arterial embolectomy, the basilar artery recanalized, and bilateral superior cerebellar arteries and posterior cerebral artery developed, although the

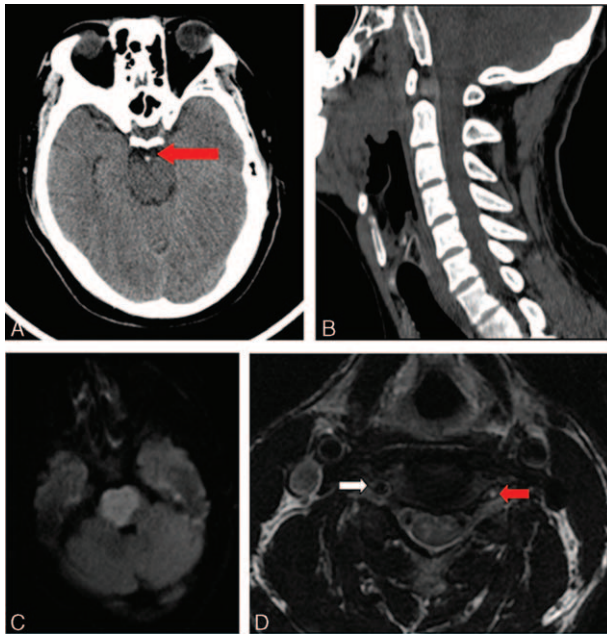


FIGURE 1. The cervical vertebra and brain CT scan and MRI scan. (A) The CT scan at 4 hours after symptoms onset showed the hyperdense basilar artery sign (the red arrow). (B) The CT scan at 4 hours after symptoms onset showed no evident abnormality in cervical cord. (C) The MRI scan at seventeenth day showed bilateral pons infarction. (D) The T2-weighted MRI image at the 17th day showed the normal right vertebral artery (white arrow) and a high-signal fulfilling the left vertebral artery (red arrow). CT = computed tomography, MRI = magnetic resonance imaging.

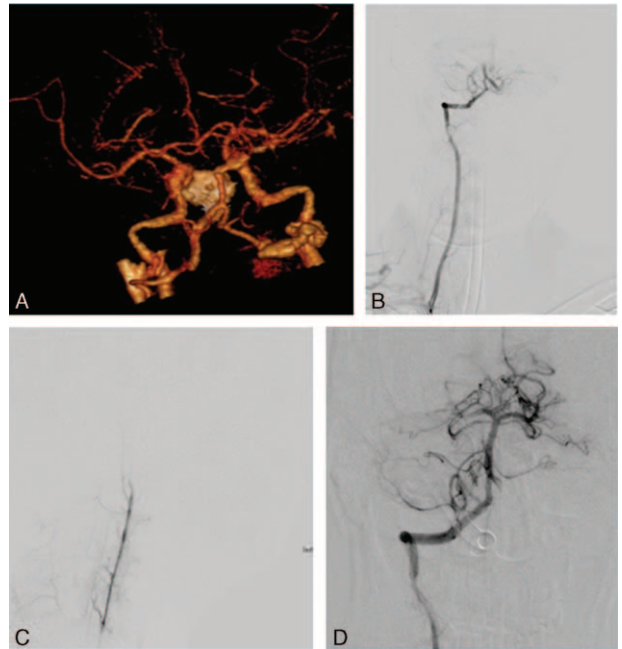


FIGURE 2. The CTA scan and DSA examination. (A) The CTA scan showed stenosis of V3 segment and part of V4 segment of left vertebral artery, stenosis of V3 segment of right vertebral artery, thrombogenesis of V4 segment of left vertebral artery, mainly blocking of distal segment of basilar artery. (B) The DSA examination showed stenosis of the left vertebral artery. (C) The DSA examination showed stenosis of the basilar artery. (D) The DSA examination after emergency embolectomy showed the basilar artery recanalized, and bilateral superior cerebellar arteries and posterior cerebral artery developed. Left vertebral artery was still completely obstructed. CTA = CT angiography, DSA = digital digital subtraction angiography.

left cerebellar artery has poor developed and still blocking of distal segment of left vertebral artery (Figure 2D). After the operation, he was returned to intensive care unit followed by antiplatelet therapy and supportive therapy. At 17 days after his admission, brain magnetic resonance imaging (MRI) scan showed pontine infarction (Figure 1C) and Axial T2-weighted showed a high-signal fulfilling the left vertebral artery (Figure 1D). Twenty-seven days later, after the treatments in the intensive care unit and a general ward, the patient's physical function gradually improved and discharged but still left neurological deficit with muscle strength grade 3/5 and hyperreflexia of limbs.

DISCUSSION

The incidence of craniocervical arterial dissection and stroke following CSM is essentially unknown. The current best available evidence suggests that craniocervical arterial dissection may be of a low incidence but could be a serious complication of CSM.⁶ In the present study, we added a rare case who was presented with LIS caused by bilateral VAD after CSM.

In 1934, a medical legal abstract 1st reported cerebrovascular accidents after CSM.⁷ In 1947, the 1st case of VAD after chiropractic manipulation was described.⁸ Dissections can be either spontaneous or traumatic. In particular, the force and extent of high velocity thrusts of the cervical spine, especially with rotational movement, can cause craniocervical arterial

dissection in susceptible individuals.⁴ Craniocervical arterial dissection is one of important causes of unpredictability of ischemic stroke in young and middle-aged people. Although current biomechanical evidence is insufficient to establish the claim that causation between CSM and craniocervical arterial dissection, clinical reports suggest that mechanical forces have a role in a considerable number of craniocervical arterial dissections and most population controlled studies have found an association between CSM and VAD stroke in young patients.⁶ Uncommonly, it has been reported bilateral VAD after CSM, resulting in the left thalamus and posterior limb of the internal capsule and the right cerebellar hemisphere infarction.⁹ In the present study, bilateral VAD after CSM causes bilateral pons infarction characterized by LIS. LIS with a vascular etiology is a particularly severe type of LIS for which the mortality is at 67% within the 1st 4 months.¹⁰

The imaging modalities are available to diagnose craniocervical arterial dissection and stroke, including duplex ultrasonography, CT, CTA, MRI, MRA, and DSA.⁶ In the present study, the cervical vertebra and brain CT scan at 1.5 hours after symptoms onset showed no evident abnormality, whereas the CT scan at 4 hours after symptoms onset showed the hyperdense basilar artery sign. The brain CTA scan showed stenosis and thrombogenesis of vertebral artery. The DSA examination confirmed stenosis of the left vertebral artery and basilar Artery. Finally, MRI scan in the 17th day confirmed bilateral pons infarction. Thus, CTA is an easy available tool to choice, and DSA is the gold standard for luminal imaging. MRI with diffusion-weighted imaging should be employed in acute infarcts and follow-up of VAD and vertebrobasilar stroke.

Up to now, no specific treatment for LIS has been established. The acute treatment and intervention of patients with LIS is similar to that for patients with other acute ischemic stroke of brain stem insults.¹¹ Currently, thrombolysis with intravenous recombinant tissue-type plasminogen activator therapy remains the only proven effective pharmacological treatment for selected acute ischemic stroke patients within a relatively short therapeutic time window of 3 to 4.5 hours after the onset of stroke symptoms,¹² although disappointments remain concerning modest recanalization rates and the devastating symptomatic intracranial hemorrhage. In 2015, the treatment protocols for acute ischemic stroke have been fundamentally altered because of the updated guidelines of American Heart Association/American Stroke Association for the early management of patients with acute ischemic stroke using endovascular techniques.¹³ This guideline concluded that certain endovascular procedures have been demonstrated to provide clinical benefit in selected patients with acute ischemic stroke. In the present study, the patient was gradually presented with LIS due to bilateral VAD, which is associated with high mortality and serious complications. Although treatment is initiated beyond 6 hours from symptom onset, we decided to proceed with a mechanical embolectomy. At result, the patient was treated by arterial embolectomy and gradually recovered to antigravity power when discharged. Thus, arterial embolectomy is one of treatment choice for vertebrobasilar thrombosis due to bilateral VAD after CSM even in later time windows.

We acknowledged that the increased risk after CSM may be an artifact of patients seeking care for neck pain resulting from existing CVD rather than the result of treatment itself,¹⁴ suggesting that not all stroke cases temporally related to CSM have preexisting craniocervical artery dissection. However, stroke is increasingly reported as a severe complication of

CSM. Thus, the safety of CSM is an issue that requires regular and rigorous assessment. The practitioner must be aware of this complication and should give the patient's informed consent to CSM. The specific radiological tests such as duplex ultrasonography and MRI should be used before CSM to evaluate the risk of adverse events and vascular vulnerability. In addition, CSM is a high-level skill of manual therapy thus necessitating training to achieve proficiency.² A well-trained practitioners may contribute to reduce the skill-related adverse events of inappropriate technique.

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