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Non responsible vascular area hyperperfusion syndrome after mechanical thrombectomy for vertebral artery occlusion: A case report

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ABSTRACT

Introduction: There are currently no published reports of hyperperfusion syndrome in the non responsible vascular area after mechanical thrombectomy for acute cerebral infarction with large vessel occlusion. Here, we report a case of hyperperfusion syndrome in the blood supply area of the right middle cerebral artery after mechanical thrombectomy for acute cerebral infarction after vertebral artery occlusion.

Patient concerns: A 21-year-old woman developed left vertebral artery occlusion, for which she received mechanical thrombectomy and successful recanalization of her occluded cerebral vessel. Subsequently, the patient became extremely agitated, with high blood pressure and headache.

Diagnosis: Two hours after the operation, bedside transcranial Doppler ultrasound examination found that the cerebral blood flow velocity of the M1 segment of the right middle cerebral artery was more than twice that of the left middle cerebral artery. Combined with the symptoms, signs and examination results of the patient, hyperperfusion syndrome in the blood supply area of the right middle cerebral artery was considered.

Interventions: The patient was administered sedation, and her pressure and ventricular rate were strictly controlled. She was no longer agitated, and her headache was significantly relieved at 36 hours after the operation.

Outcomes: On the 5th day after the operation, the blood flow velocity of her right middle cerebral artery decreased to normal level, and the patient recovered well.

Conclusion: In this case, after mechanical thrombectomy, such patients with acute posterior circulation cerebral infarction can experience hyperperfusion syndrome in the non responsible vascular area of the anterior circulation. Bedside transcranial Doppler cerebral blood flow examination can identify the hyperperfusion state of cerebral vessels in a timely manner and effectively guide treatment.

Abbreviations: NIHSS, National Institute of Health Stroke Scale; GCS, Glasgow Coma Score; CT, computed tomography; MRI, magnetic resonance imaging; MRA, magnetic resonance angiography; PI, pulse index; RI, resistance index; CTA, computed tomographic angiography.

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1. Introduction

Cerebral hyperperfusion syndrome refers to an increase in cerebral blood flow exceeding the demand of brain tissue, resulting in various clinical symptoms [1]. Hyperperfusion syndrome can be seen after carotid stent implantation or carotid endarterectomy. The overall incidence of hyperperfusion syndrome of patients after treated with after carotid endarterectomy or carotid stent implantation was about 1.4% [2]. The risk factors for hyperperfusion syndrome include severe stenosis of the cerebral artery, insufficient compensatory capacity of the collateral vessels, hypoperfusion in the distal arterial supply area and so on [3]. A small number of studies [4–6] have reported that hyperperfusion syndrome in the responsible vascular area can also occur after mechanical

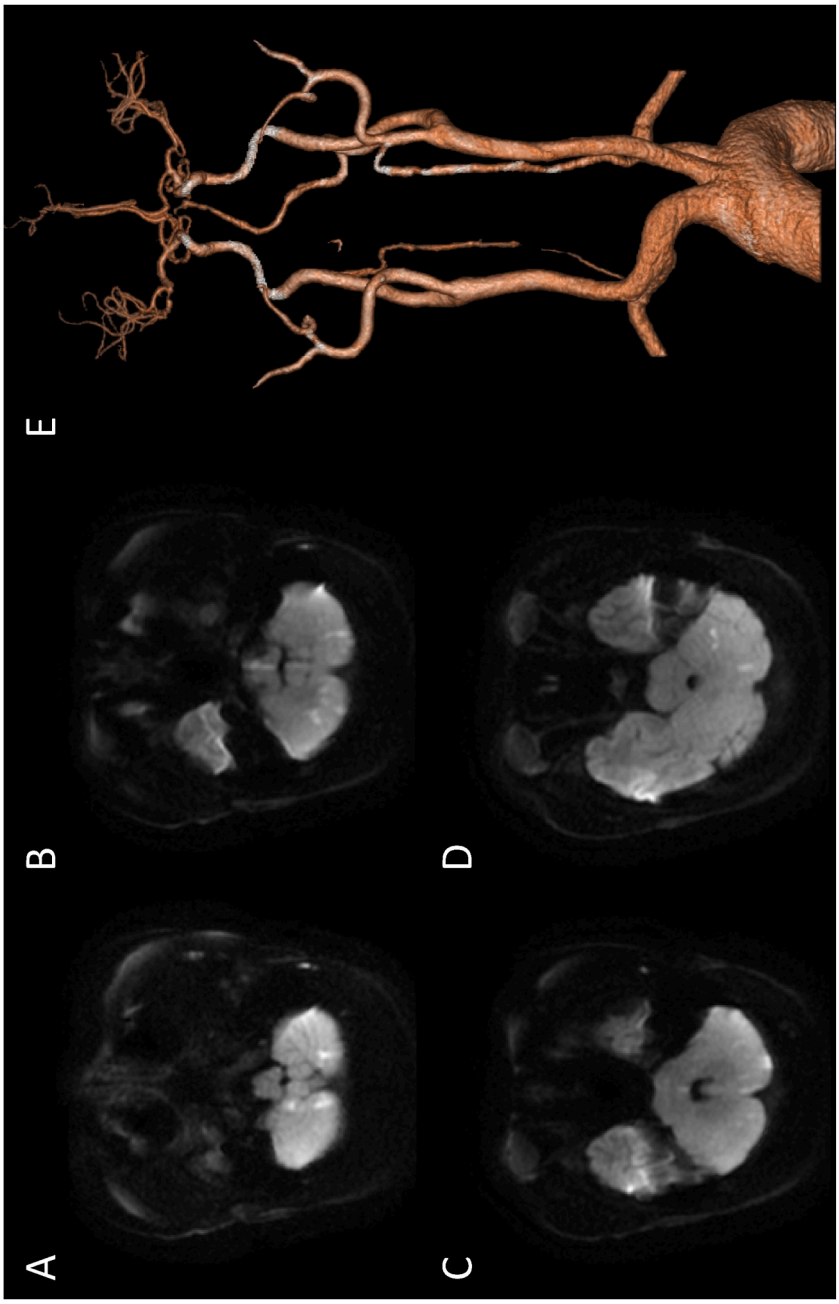

Fig. 1. Preoperative brain MRI showed multiple acute lacunar infarctions in the bilateral cerebellar hemisphere and pons (1A and B). On the second day after the operation, brain MRI examination showed acute infarction in the bilateral cerebellar hemisphere, pons and medulla oblongata (1C and D). On the second day after the operation, head and neck CTA showed mild stenosis of the V3 segment of the left vertebral artery and severe local stenosis of the M2 segment of the right middle cerebral artery (1E).

thrombectomy for acute cerebral infarction with large vessel occlusion. However, there has never been a report of hyperperfusion syndrome in the non responsible vascular area after mechanical thrombectomy for acute cerebral infarction with large vessel occlusion. This paper reports a case of hyperperfusion syndrome in the blood supply area of the right middle cerebral artery after mechanical thrombectomy for acute cerebral infarction subsequent to vertebral artery occlusion.

2. Case presentation

The patient was a 21-year-old woman who had a history of type 1 diabetes for 3 years and hypertension for 2 years. She did not use medication regularly, and her blood sugar and blood pressure were not well controlled. This time, due to dizziness for 1 week, weakness of her right limb and unclear speech for more than 5 hours, she was sent to the emergency department of our hospital for treatment. Physical examination showed clear consciousness, unclear speech, a shallow nasolabial groove on the right side, grade-2 muscle strength of the right upper limb, grade-3 muscle strength of the right lower limb, grade-5 muscle strength of the left limb, shallow hypoesthesia of the right limb, positive pathological signs on the right side, National Institute of Health Stroke Scale (NIHSS) was 8 points, and Glasgow Coma Score (GCS) was 14 points. No hemorrhage was found in the emergency brain computed tomography (CT) examination, and the aggravation time of the patient's symptoms was less than 6 hours. The patient was given intravenous thrombolysis with 1 million units of urokinase. After thrombolysis, the patient's symptoms were not significantly relieved, and large vessel occlusion was not ruled out. Urgent brain magnetic resonance imaging (MRI) examination suggested multiple acute lacunar infarctions in the bilateral cerebellar hemisphere and pons (Fig. 1A and B). The patient was extremely agitated and repeatedly received diazepam injection as well as midazolam injection but was still unable to cooperate with magnetic resonance angiography (MRA)

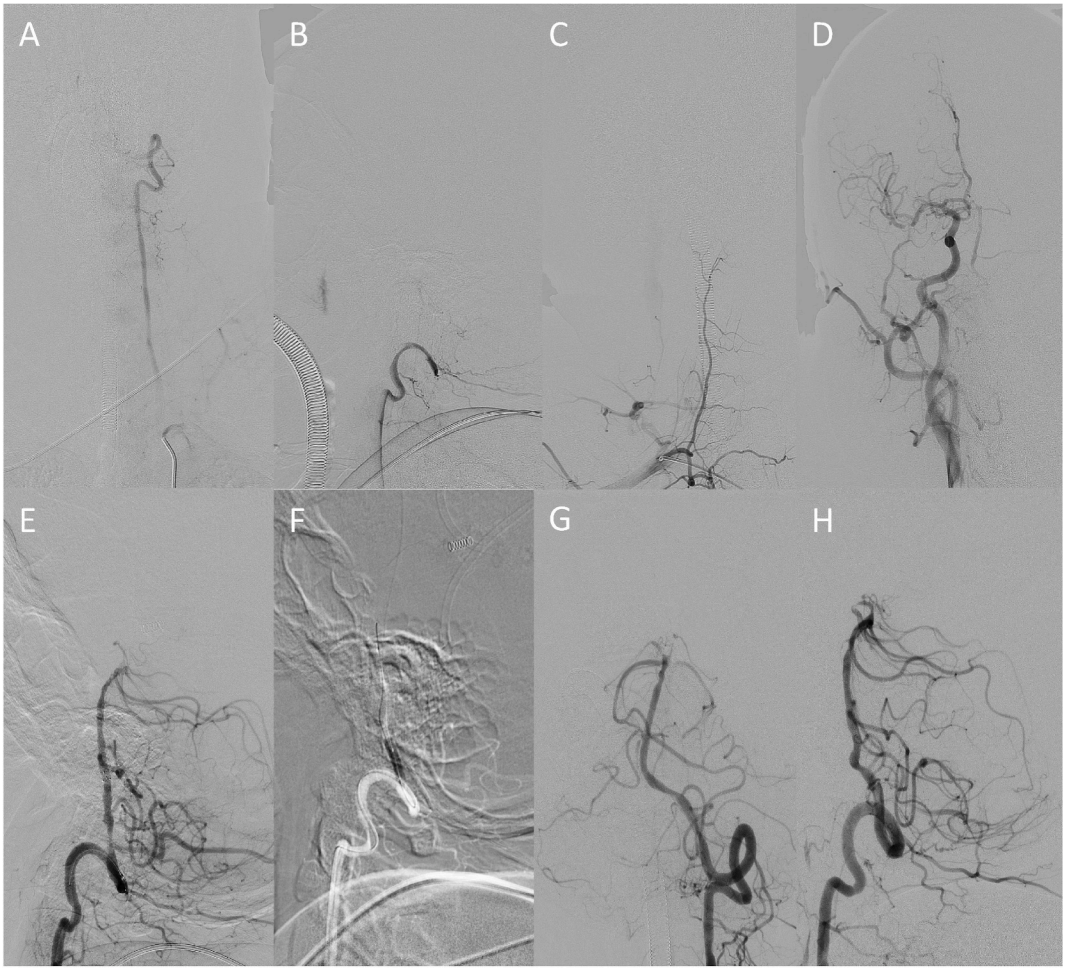

Fig. 2. Cerebral angiography showed occlusion of the beginning of the right vertebral artery, occlusion of the V3 segment of the left vertebral artery, and severe local stenosis of the M2 segment of the right middle cerebral artery (2A, B, C and D). Stent thrombus removal of the V3 segment of the left vertebral artery was performed (2E). Balloon dilatation was performed at the point of severe stenosis in the V3 segment of the left vertebral artery (2F). Cerebral angiography showed mild stenosis of the V3 segment of the left vertebral artery and no obvious thromboembolism at the distal end (2G and H).

examination to further evaluate the cerebrovascular situation. According to the symptoms and signs of the patient, we considered the high possibility of posterior circulation large vessel occlusion. There were indications for emergency cerebral angiography to confirm whether vessels were occluded and to perform a mechanical thrombectomy when necessary. The family members signed their consent and agreed to the application of emergency mechanical thrombectomy if necessary. The patient's preoperative blood pressure was 188/107 mmHg, and her pulse was 126 times/min. Protective endotracheal intubation was urgently performed. Cerebral angiography under local anaesthesia plus midazolam and dexmedetomidine sedation showed occlusion of the beginning of the right vertebral artery, occlusion of the V3 segment of the left vertebral artery, local severe stenosis of the M2 segment of the right middle cerebral artery, and opening of the right posterior communicating artery (Fig. 2A, B, C and D). According to the results of cerebral angiography, our team quickly discussed and then performed stent removal of the thrombus of the left vertebral artery V3 (Fig. 2E). The stent we used was a solitaire AB (6 × 30 mm). After stent removal of the thrombus, angiography showed severe local stenosis of the left vertebral artery V3. Next, a balloon dilation catheter (Gateway 3.0 × 15 mm) was used to dilate the severe stenosis of the V3 segment of the left vertebral artery with 6 atmospheric pressures (Fig. 2F). After dilation, angiography showed mild stenosis of the V3 segment of the left vertebral artery. To prevent blood vessel from occluding again, a total of 6 ml of tirofiban hydrochloride injection was injected at a uniform rate of 1 ml/min along the 8F catheter. After waiting for 15 minutes, cerebral angiography showed mild stenosis of the V3 segment of the left vertebral artery and no obvious thromboembolism at the distal end (Fig. 2G and H). The patient's blood pressure was 125/84 mmHg when leaving the operation room. After the operation, no hemorrhage was found on brain CT. After returning to the ward, the patient became extremely agitated, her blood pressure fluctuated between 181/101 and 182/103 mmHg, and her ventricular rate was 150–160 times/min. The examination of the nervous system of the patient showed that the patient could pronounce, the diameter of the pupils on both sides was about 2.5 mm, which was sensitive to light reflex directly and indirectly. The muscular strength of the right limb was roughly detected as Grade 4, and the pathological signs on the right side were positive. Two hours after the operation, the bedside transcranial Doppler ultrasound showed that the blood flow velocity of the M1 segment of the

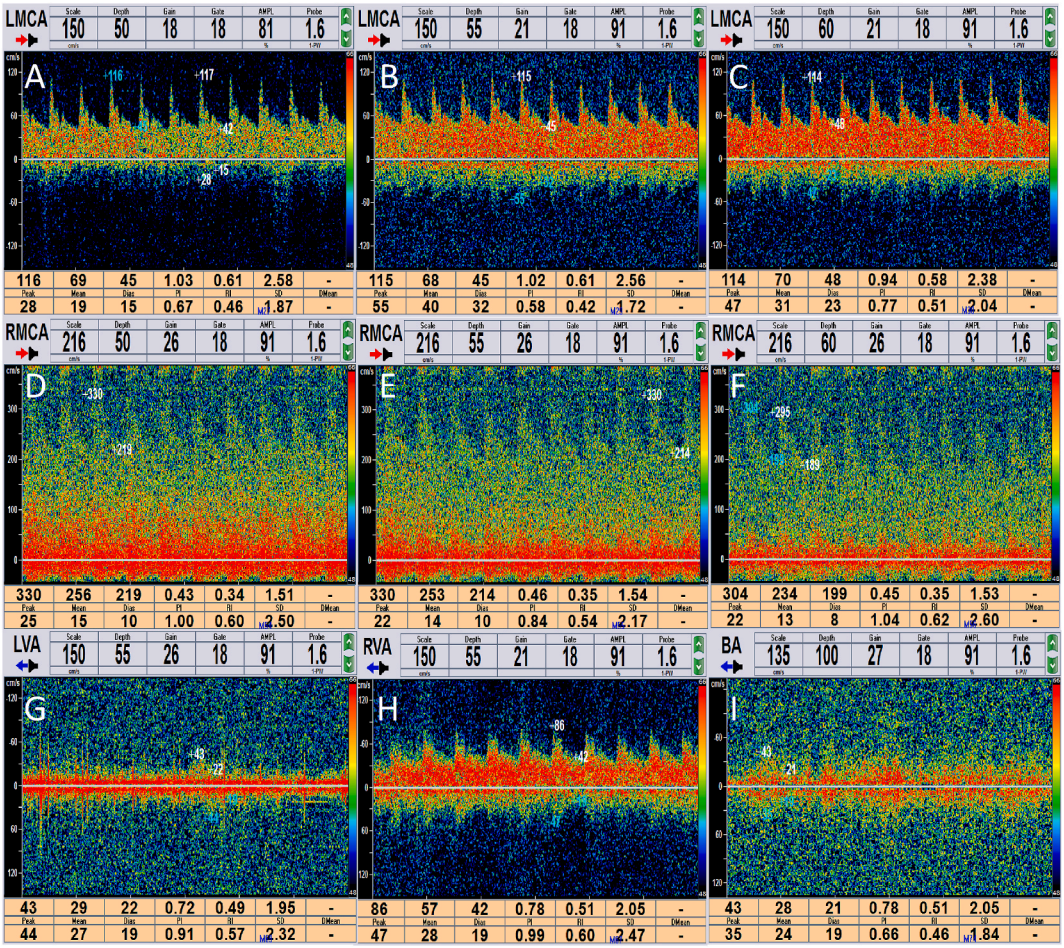

Fig. 3. Two hours after the operation, bedside transcranial Doppler ultrasound showed that the blood flow velocity of the M1 segment of the right middle cerebral artery fluctuated between 304 and 330 cm/s **(3D, E and F)**, the blood flow velocity of the M1 segment of the left middle cerebral artery fluctuated between 114 and 117 cm/s **(3A, B and C),** and the blood flow velocities of the bilateral vertebral artery and basilar artery were in the normal range **(3G, H and I)**. The PI and RI values of all vessels were in the normal range **(3A-I)**.

right middle cerebral artery fluctuated between 304 and 330 cm/s, that of the M1 segment of the left middle cerebral artery fluctuated between 114 and 117 cm/s, and that of the bilateral vertebral arteries and basilar artery were in the normal range. The pulse index (PI) and resistance index (RI) values of all cerebral vessels were in the normal range (Fig. 3), which did not support the acceleration of the blood flow velocity of the right middle cerebral artery caused by cerebrovascular stenosis or cerebral vasospasm. Combined with the fact that the blood flow velocity of the right middle cerebral artery was more than twice that of the left middle cerebral artery, the blood pressure of the patient was higher than 180/100 mmHg within 2 hours after the operation, and the ventricular rate was greater than 100 times/min and considering the existence of hyperperfusion syndrome in the blood supply area of the right middle cerebral artery, after full discussion, our team decided to use nicardipine to actively control the blood pressure below 140/90 mmHg and continued to administer midazolam injection for sedation and β receptor blockers to control the patient's ventricular rate and avoid possible symptoms of hyperperfusion syndrome, such as intracerebral hemorrhage and seizures. In terms of antiplatelet drugs, tirofiban injection was injected intravenously for 24 hours, and then aspirin and clopidogrel were taken orally after 24 hours. After stopping sedative drugs 24 hours after the operation, the patient was drowsy, extremely agitated and complained of headache. No hemorrhage or edma was found on brain CT. Sedation, blood pressure control and ventricular rate control were continued. At the 36th hour after the operation, the patient's blood pressure was 132/80 mmHg, the ventricular rate fluctuated between 100 and 120 beats/min, sedative drugs were stopped, the patient regained clear consciousness, her headache was significantly relieved, and she was no longer agitated. The patient exhibited spontaneous breathing and a good cough reflex, and the endotracheal tube was removed. The physical examination showed that the patient's speech was basically clear, the muscle strength of her right limb had recovered to grade 4, and the NIHSS score decreased to 3 points. On the second day after the operation, brain MRI reexamination showed acute infarction in the bilateral cerebellar hemisphere, pons and medulla oblongata (Fig. 1C and D). At the same time, no brain edema or new infarction lesions were found in other parts of the brain. Head and neck computed tomographic angiography (CTA) showed mild stenosis of the V3 segment of the left vertebral artery and local severe stenosis of the M2 segment of the right middle cerebral artery (Fig. 1E). On the 5th day after the operation, transcranial Doppler ultrasonography showed that the blood flow velocity of the right middle cerebral

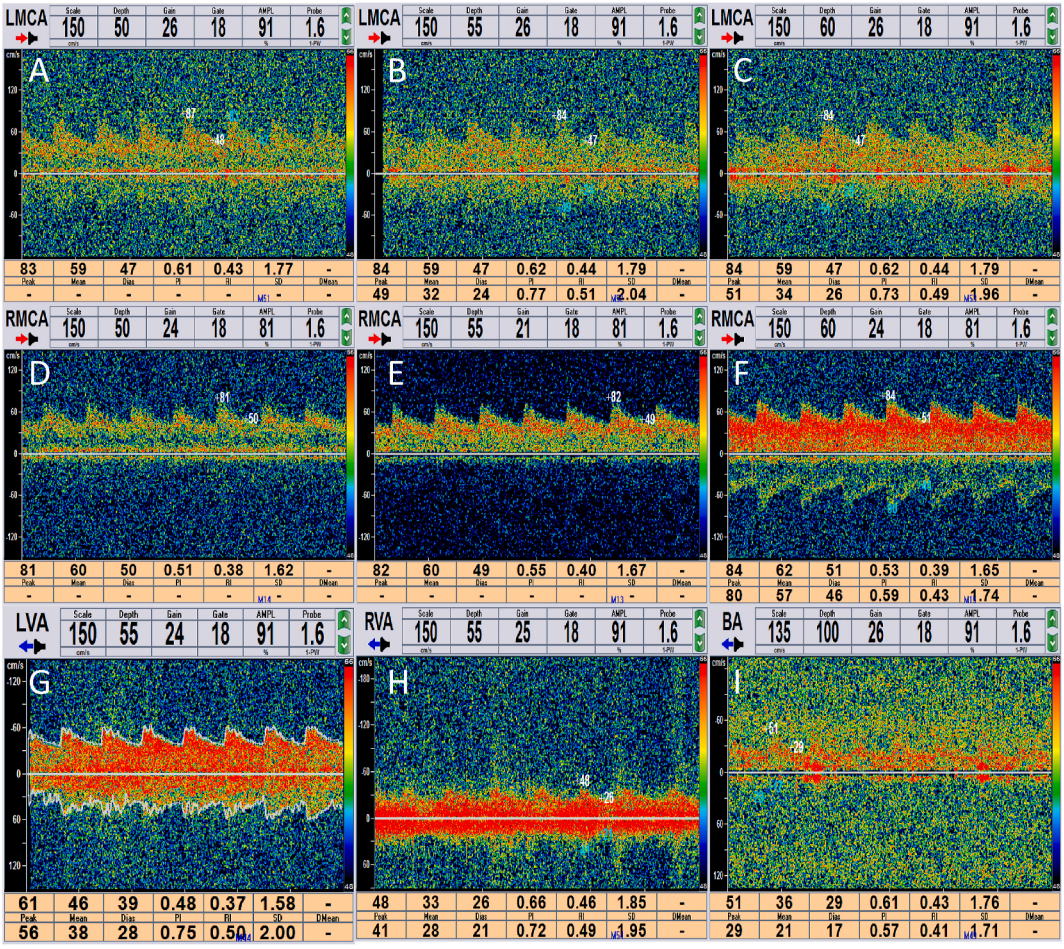

Fig. 4. On the 5th day after the operation, transcranial Doppler ultrasonography showed that the blood flow velocity of the right middle cerebral artery fluctuated from 81 to 84 cm/s **(4D, E and F)**, which was basically symmetrical with the blood flow velocity of the left middle cerebral artery **(4A, B and C)**, and the blood flow velocity, PI value and RI value of all cerebral vessels were in the normal range **(4A-I)**.

artery fluctuated from 81 to 84 cm/s, which was basically symmetrical with that of the left middle cerebral artery. The blood flow velocities of the left middle cerebral artery, bilateral vertebral arteries, and basilar arteries were in the normal range. The PI and RI values of all cerebral vessels were in the normal range (Fig. 4). Considering that the hyperperfusion syndrome of the right middle cerebral artery had been relieved, we continued to control the patient's blood pressure at or below 140/90 mmHg, and the patient showed no signs of agitation or new neurological deficits. The patient's symptoms improved significantly one week after onset, and she was discharged from the hospital. The NIHSS score was 2 at the time of discharge, and the mRS score was 1 after 3 months of follow-up, which meant that the prognosis of the patient was good.

3. Discussion

Cerebral hyperperfusion syndrome refers to an increase in cerebral blood flow exceeding the demand of brain tissue, resulting in various clinical symptoms [1]. Cerebral hyperperfusion syndrome has been widely reported after carotid endarterectomy or carotid stent implantation [2,3], but there are few literature reports of hyperperfusion syndrome after mechanical thrombectomy for acute cerebral infarction with large vessel occlusion. In 2007, Ogasawara et al. retrospectively analysed 4494 patients with carotid stenosis treated with carotid endarterectomy or carotid stent implantation and found that the overall incidence of hyperperfusion syndrome was 1.4% [2]. Shimonaga K et al. [4] reported that 13 cases of hyperperfusion were found among 27 patients receiving successful mechanical thrombectomy for acute cerebral infarction. The incidence of hyperperfusion after thrombectomy reported by Shimonaga K et al. [4] was 48.15%, which was high and could not represent the actual incidence of hyperperfusion syndrome after mechanical thrombectomy in patients with acute cerebral infarction.

Hyperperfusion syndrome can manifest as severe headache, extreme agitation, seizures, focal neurological dysfunction, visual impairment, intracranial hemorrhage and other conditions [1]. Our patient was extremely agitated, with high blood pressure and a fast ventricular rate, which was in line with the clinical manifestation of hyperperfusion syndrome.

The risk factors for hyperperfusion syndrome include severe stenosis of the cerebral artery, insufficient compensatory capacity of the collateral vessels, hypoperfusion in the distal arterial supply area, decreased cerebral reserve capacity, increased regional cerebral blood flow before surgery, history of hypertension, and history of diabetes mellitus [3]. It has been reported that hyperperfusion syndrome after mechanical thrombectomy for acute cerebral infarction caused by cardiogenic embolism is related to the slow generation of collateral circulation [5]. Ye J et al. [5] reported a patient with regional hyperperfusion syndrome in responsible vessels after mechanical thrombectomy for cardiogenic cerebral embolism. Transcranial Doppler ultrasonography showed that the blood flow velocity of the M1 segment of the right middle cerebral artery increased significantly. After enhanced hypotension, the blood flow velocity of the right middle cerebral artery returned to normal, and the prognosis of the patient was good. It is believed that the primary pathogenesis of hyperperfusion syndrome includes the impairment of brain autonomic regulation function, and other pathogeneses include the formation of reactive oxygen species, continuous injury of cerebrovascular endothelial cells, destruction of the baroreceptor reflex and so on. Kneihsl M et al. [6] reported a patient after mechanical thrombectomy for left internal carotid artery occlusion. Transcranial Doppler examination revealed that the blood flow velocity of the left middle cerebral artery had increased significantly, and the patient had involuntary twitching of the right upper limb. Electroencephalograph examination showed that there were intermittent sharp and slow complex waves. After intensive antihypertensive and antiepileptic treatment, the patient's consciousness gradually recovered, but the paralysis was serious, and the prognosis was poor. Our patient had a history of type 1 diabetes and hypertension, without sufficient blood-pressure and blood-sugar control. The blood velocity of the M1 segment of the right middle cerebral artery was markedly high after mechanical thrombectomy, and the M2 segment of the right middle cerebral artery was severely stenosed, which suggested that the vascular bed of the right middle cerebral artery was relatively weak and that the cerebral blood flow automatic regulation ability was poor.

Before the V3 segment of the occluded left vertebral artery was opened, the right anterior circulation provided a compensatory blood supply to the posterior circulation through the opened right posterior communicating artery. After the V3 segment of the occluded left vertebral artery was opened, the blood supply of the posterior circulation was quickly restored, and the demand for the compensatory effect of the posterior circulation on the cerebral blood flow of the right anterior circulation was reduced. Two hours after the operation, transcranial Doppler examination showed that the cerebral blood flow velocity of the M1 segment of the right middle cerebral artery was more than twice that of the left middle cerebral artery. The cerebral blood flow supplied to the right middle cerebral artery may have far exceeded its actual needs for cerebral blood flow. At the same time, it was not found that the blood flow of the right middle cerebral artery was diverted to other large blood vessel supply areas, and the collected transcranial Doppler ultrasound examination information did not reveal the basis for cerebral vasospasm or severe stenosis in the M1 segment of the middle cerebral artery. After sedation and strict control of blood pressure, the transcranial Doppler ultrasound showed that the blood flow velocity of the right middle cerebral artery decreased to a value indicating symmetry with the left middle cerebral artery. The brain MRI examination did not reveal any new cerebral infarction lesions in the right cerebral hemisphere, which was in line with the diagnostic criteria of hyperperfusion syndrome formulated by Bouri et al. [7].

This patient presented with agitation, headache and other symptoms after mechanical thrombectomy, which should be differentiated from hypertensive encephalopathy and cerebral ischemia reperfusion injury. Hypertensive encephalopathy refers to the increase of blood pressure leading to cerebral vascular sclerosis and spasm, which leads to the aggravation of brain edema [8]. Transcranial Doppler ultrasound examination 2 h after the operation of this patient did not find sclerosis or spasm in M1 segment of the right middle cerebral artery or other cerebral vessels, which could exclude the diagnosis of hypertensive encephalopathy. The most typical manifestations of cerebral ischemia reperfusion injury are brain edema and brain cell necrosis [9]. Firstly, in this case, brain CT examination immediately after mechanical thrombectomy did not find brain edema in the right hemisphere. Secondly, brain MRI

reexamination on the second day after mechanical thrombectomy did not find obvious brain edema or new cerebral infarction lesions in the anterior circulation. Thirdly, the infarction area of brain MRI reexamination in the posterior circulation did not significantly increase compared with that before thrombectomy. According to the above three points, we confirmed that the diagnosis of cerebral ischemia reperfusion injury can be ruled out.

The vessel responsible for this patient's ischemic stroke was the left vertebral artery. After mechanical thrombectomy of the left vertebral artery, hyperperfusion syndrome appeared in the blood supply area of the right middle cerebral artery of the non responsible vessel, suggesting that hyperperfusion syndrome can also appear in the blood supply area of the non responsible vessel after mechanical thrombectomy performed for acute cerebral infarction. Bedside transcranial Doppler cerebral blood flow examination can be used to determine whether there is hyperperfusion syndrome in patients with acute cerebral infarction after mechanical thrombectomy in a timely manner and can effectively guide the regulation of blood pressure and whether to continue to use sedative drugs to avoid the occurrence of epilepsy, cerebral hemorrhage and other complications that may occur in patients with hyperperfusion syndrome [10,11].

In conclusion, in this case, after mechanical thrombectomy, such patients with acute cerebral infarction of the posterior circulation can experience hyperperfusion syndrome in the non responsible vascular area of the anterior circulation. Bedside transcranial Doppler cerebral blood flow examination can identify the hyperperfusion state of cerebral vessels in a timely manner and effectively guide treatment.

Conflicts of interest and source of funding

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Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

Data availability statement

Data included in article/supp. material/referenced in article.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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