

# Delayed hemorrhage after microvascular decompression in a hemifacial spasm patient associated with neurosyphilis

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## Abstract

Microvascular decompression is an effective treatment for hemifacial spasm. Hemorrhage is one of the complications of microvascular decompression. However, delayed hemorrhage is very rare. Here, we report a case of ipsilateral cerebellar hemorrhage at day seven following microvascular decompression. A 45-year-old woman presented with left HFS for the previous two years. Diagnostic testing demonstrated the presence of neurosyphilis. Brain magnetic resonance image was unremarkable on presentation. She received microvascular decompression and her hemifacial spasm completely resolved after surgery. At day seven post-operatively, the patient presented with a sudden onset headache. Emergency computed tomography scan showed a cerebellar hemorrhage. A suboccipital craniotomy was performed and a cerebellar hematoma was evacuated. The delayed hemorrhage was attributed to possible microaneurysm from syphilis.

**Keywords:** Hemifacial spasm; microvascular decompression; neurosyphilis; delayed hemorrhage

## INTRODUCTION

Hemifacial spasm (HFS) is characterized by involuntary, unilateral, irregular clonic or tonic movements of the muscles, innervated by the ipsilateral facial nerve, usually beginning around the eyes before progressing inferiorly to the cheek, mouth, and neck.<sup>1</sup> The prevalence of HFS is 9.8 per 100,000.<sup>2</sup> Primary HFS is the result of vascular compression at the facial nerve root entry zone in the posterior fossa.<sup>3, 4</sup> Secondary HFS is due to facial nerve compression produced by tumors, demyelinating diseases, and a range of other causes.<sup>5</sup>

Microvascular decompression (MVD) is a curative treatment for primary HFS.<sup>6</sup> Hemorrhage is one of the complications of MVD, but delayed hemorrhage is scarcely seen. Herein, we report a case of a patient with neurosyphilis who underwent MVD for left HFS, and developed an ipsilateral cerebellar hemorrhage on the seventh postoperative day.

## CASE REPORT

A 45-year-old woman complained of a history of intermittent, involuntary muscle spasms involving

the left side of her face. These symptoms began two years earlier. Initially only the external angle of the eye was affected. Then, the ipsilateral lower facial muscles were subsequently involved. The intensity and frequency of spasms gradually increased over the past six months. Initially, she was treated with carbamazepine, then botulinum toxin injections with minimal symptomatic improvement.

On admission to our hospital, she had involuntary, paroxysmal contractions of the facial muscles on the left side, provoked during talking and when anxious. Neurological examination showed normal bilateral corneal and facial sensation, and intact hearing. There was no facial weakness.

A routine blood test showed she was positive for the presence of *Treponema pallidum* (*Spirochaeta pallida*) antibody. Further serum *Treponema pallidum* particle agglutination (TPPA) and rapid plasma regain (RPR) tests were also positive. She denied any history of genital ulcers, which can be caused by the infection of *T. pallidum*. Syphilis can affect virtually any organ of the human body.<sup>7</sup>

Lumbar puncture was performed and the analysis of cerebrospinal fluid (CSF) revealed a

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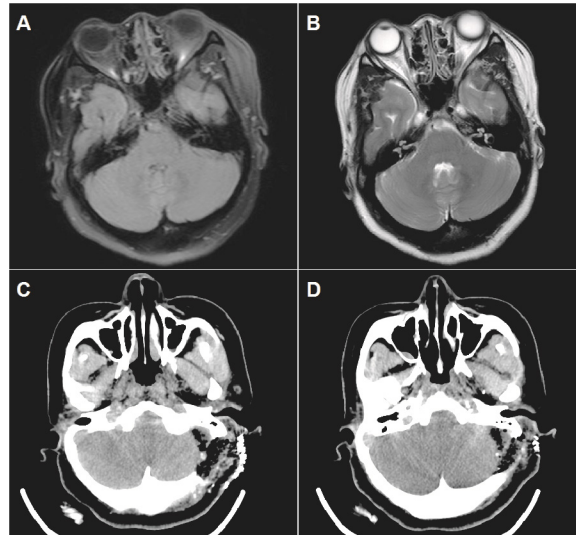


Figure 1. The cerebellopontine angle (CPA) was normal on axial T1-(A) and T2-(B) weighted MRI before microvascular decompression (MVD). CT showed no hemorrhage immediately after MVD (C-D).

protein level of 0.39 g/L (normal range 0.15–0.45 g/L), a glucose level of 3.13 mmol/L (normal range 2.3–4.3 mmol/L). CSF *T. pallidum* antibody, and CSF RPR and TPPA were all positive.

Magnetic Resonance Imaging (MRI) (Figure 1 A,B) and computed tomography (CT) scans after admission to our hospital showed no obvious abnormalities in her brain. She was diagnosed with primary HFS with concurrent neurosyphilis and received penicillin (24x10<sup>6</sup> IE IV/day, followed by 2.4x10<sup>6</sup> units, intramuscular, once weekly for three weeks) for 14 days. She was subsequently referred to our neurosurgical team.

Standard retromastoid suboccipital craniotomy and MVD were performed. Teflon was interposed

between the posterior inferior cerebellar arteries and the facial nerve root entry zone region (Figure 2A,B,C). After surgery, her symptoms of hemifacial spasm completely resolved and a CT scan showed no hemorrhage present (Figure 1C,D).

During the first six days after surgery, the patient had a non-eventful recovery from surgery. Blood pressure is in normal range. However, on the seventh postoperative day, she suddenly complained of a headache. Neurological examination showed a decreased level of consciousness. Blood pressure was 180/118mmHg and pulse rate was 85 beats per minute. The patient was afebrile and comatose (Glasgow Coma Scale

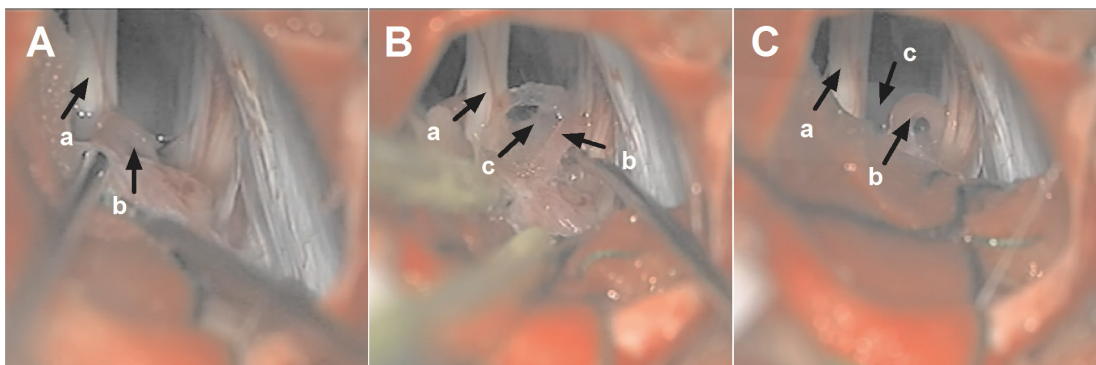


Figure 2. (A) Before the microvascular decompression (MVD), the facial nerve (a) was compressed by posterior inferior cerebellar artery (PICA) (b) at nerve root entry zone in the posterior fossa; (B) Teflon (c) was interposed between the PICA (b) and the facial nerve (a) at nerve root entry zone; (C) After MVD, the facial nerve (a) was not compressed by PICA (b). Teflon (c) was interposed between the PICA (b) and the facial nerve (a).

9/15). Neurological examination showed that the pupil was equal and reactive to light. There was cerebellar ataxia and right hemiparesis. Both Babinski signs are negative. A brain computed tomography (CT) scan demonstrated left cerebellar hemorrhage with mass effect (Figure 3A,B).

The patient was treated with hydrochlorothiazide 12.5 mg daily, and mannitol 250 mL twice a day. One day later, head CT angiogram showed no aneurysms in the cerebellum (Figure 3C,D). The level of consciousness of the patient deteriorated with Glasgow Coma Scale of 7/15. A sub-occipital craniotomy was performed followed by cerebellar hematoma evacuation (Figure 3E,F). The patient's general condition as well as mental status improved significantly following surgery. She was discharged on the tenth day after sub-occipital craniotomy. At 2 months follow-up, the patient was conscious and orientated. Her speech was fluent. The neurological function of both limbs were normal.

## DISCUSSION

MVD also is considered first choice for the primary surgical treatment of HFS. In China such

a procedure is very popular and widely adopted.<sup>8-11</sup>

MVD is an effective and safe treatment option for HFS. While MVD surgery may cause some complications such as facial paralysis,<sup>12</sup> hemorrhage is rare,<sup>13</sup> and usually occurs within the first few hours after operation.<sup>14,15</sup> Delayed hemorrhage is very uncommon.<sup>16</sup> We reviewed the literature on delayed hemorrhage after MVD for HFS as listed in Table 1.

The PubMed and Web of Science were searched independently by two reviewers using following keywords and subject terms: "delayed hemorrhage" in combination with "microvascular decompression". The last search was on November 1, 2016.

Although the exact cause of delayed hemorrhage after MVD cannot be confirmed, some explanations have been proposed to account for delayed hemorrhage. Our literature review shows that MVD-associated delayed hemorrhage is most often caused by an aneurysm,<sup>13,17,18</sup> arteriovenous fistula.<sup>19</sup> Hanakita *et al.*<sup>13</sup> reported a patient who suffered with a delayed hemorrhage after MVD due to a traumatic aneurysm. Li *et al.*<sup>16</sup> suggested that excessive cerebrospinal fluid (CSF) drainage causing laceration of the bridging veins, and

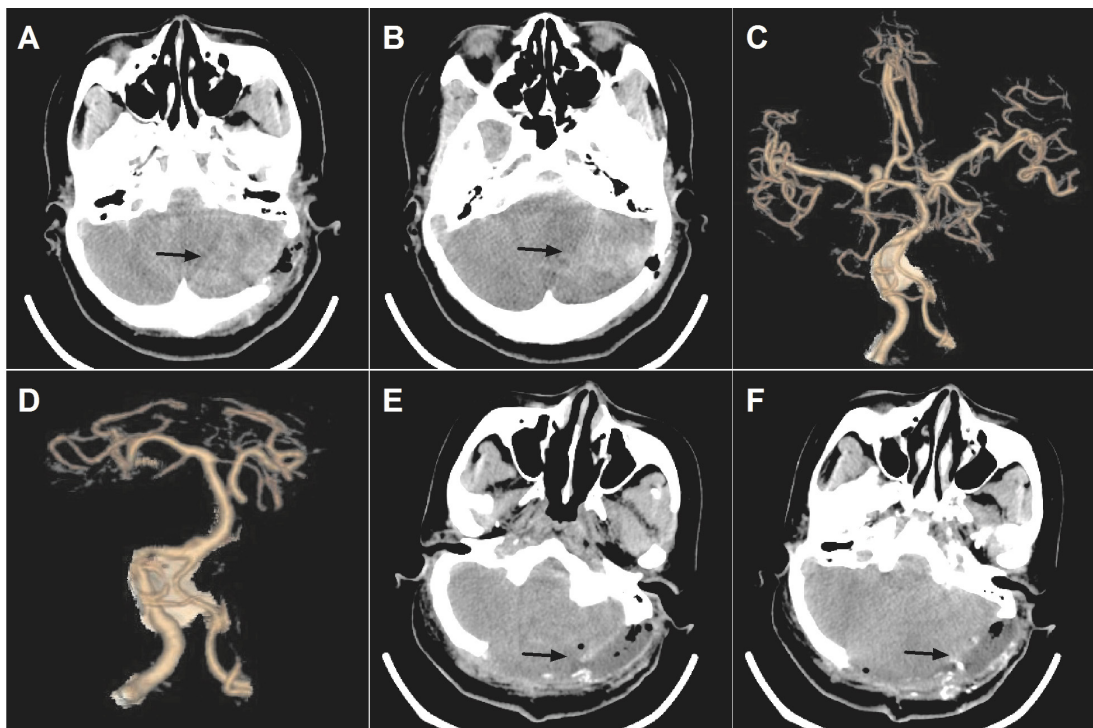


Figure 3: Multi-slice CT images show left cerebellar hemorrhage (arrow) at seventh day post microvascular decompression (A-B). CT angiography shows normal vasculature in the whole brain (C) and in the posterior brain (D). Multi-slice CT images show suboccipital craniotomy (arrow) and cerebellar hematoma evacuation (E-F).

**Table 1: Previous reports of delayed hemorrhage of MVD**

Author	Case	Age	Sex	HFS Location	Cause	Time of delayed hemorrhage	Complication	Treatment	Outcome
Kim <i>et al.</i> <sup>19</sup>	1	49	M	Left side	AICA	10 months PO	Dural AVF of the left transverse sigmoid sinus	Embolization with Onyx® on AVF	Discharged with no deficit
Li <i>et al.</i> <sup>16</sup>	2	70	M	Left side	PICA	3 <sup>rd</sup> day PO	Subdural hematoma in the left CPA	Surgery to partially remove the hematoma	Dead
	3	60	F	Left side	AICA	14 <sup>th</sup> day PO	Subdural hematoma in the left CPA	None	Discharged with no deficit
	4	47	F	Right side	—	2 <sup>nd</sup> day PO	Hemorrhage in the left parietal and temporal lobes	Surgery to remove the hematoma	Discharged with slight aphasia
Kudo <i>et al.</i> <sup>17</sup>	5	39	F	Left side	PICA	1 year PO	A dissecting aneurysm of the left vertebral artery	Endovascular parent artery occlusion of the dissecting aneurysm	Discharged with moderate disability
Tokuda <i>et al.</i> <sup>18</sup>	6	44	F	Right side	AICA	9 years PO	Subarachnoid hemorrhage caused by the rupture of aneurysm of AICA	Clipped 2 days after onset of the subarachnoid hemorrhage	Discharged
Hanakita <i>et al.</i> <sup>13</sup>	7	51	F	Right side	—	10 <sup>th</sup> day PO	Subarachnoid hemorrhage due to aneurysm in the right AICA	Clipped at emergency craniotomy	Discharged with no deficit

F = female; M = male; AICA= anterior inferior cerebellar artery; PICA= posterior inferior cerebellar artery; AVF= arteriovenous fistula; CPA= cerebellopontine angle; — = not available; PO= postoperative

retraction of the cerebellum, flexion of the neck and excessive rotation of the head may also result in delayed hemorrhage. Some investigators<sup>17,18,20</sup> have reported cases of delayed hemorrhage due to ethyl 2-cyanoacrylate, used to fix the compressing vessel to the dura mater in surgery, resulting in aneurysms. Kim *et al.*<sup>19</sup> suggested that craniotomy may cause acquired dural arteriovenous fistula (AVF) of the transverse-sigmoid sinus.

Our team drained CSF slowly, operated cautiously, and did not use ethyl 2-cyanoacrylate during surgery. A CT angiogram (CTA) after delayed hemorrhage did not show any arteriovenous malformations (AVM) or aneurysms. Our patient's blood pressure was also within the normal range.

Our patient had syphilis, and was not aware of the infection before presentation to our hospital. Invasion of the central nervous system by spirochetes may occur at any time in the course of syphilis in up to 40% of untreated patients,<sup>21</sup> manifesting as syphilitic meningitis, meningovascular syphilis, parenchymatous neurosyphilis, and gummatous neurosyphilis.<sup>22</sup> Some patients with neurosyphilis are asymptomatic, with evidence of syphilis only in serological testing or CSF analysis.<sup>23</sup> In our patient, the diagnosis of neurosyphilis was confirmed by the positive RPR and TPPA tests in both blood and CSF.

During the early stages of neurosyphilis, the meninges and vasculature are most commonly affected.<sup>7</sup> Some investigators have reported that neurosyphilis may result in the development of microaneurysm in the brain.<sup>24</sup> Vascular damage leads to fibroblastic proliferation of the intima, thinning of the media, and fibrous and inflammatory abnormalities in the adventitia, with lymphocytic and plasma cell infiltration, leading to formation of aneurysms.<sup>24</sup> As the patient refused CTA and MR angiography before MVD, we do not know whether any vascular abnormalities were present prior to MVD. Although CTA after MVD showed no evidence of aneurysms, the possibility of a microaneurysm cannot be excluded and may have ruptured and resulted in hemorrhage. Therefore, our patient may have had a microaneurysm that led to delayed hemorrhage.

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This manuscript has been edited and proofread by Medjaden Bioscience Limited. The study was approved by the ethics committee of First Hospital of Jilin University. Informed parental consent was obtained in this case. We have received a signed

release form from the patient parents authorizing the publication of her material.

## DISCLOSURE

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Conflict of interest: None

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