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Case Report

Coil Embolization of VA-PICA Aneurysm Presenting with Hemifacial Spasm with Assistance of Abnormal Muscle Response Monitoring

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Objective: We report a rare case of symptomatic vertebral and posterior inferior cerebellar arteries (VA-PICA) aneurysm-caused ipsilateral hemifacial spasm (HFS) for which coil embolization of the aneurysm with the assistance of abnormal muscle response (AMR) monitoring was effective.

Case Presentation: A 62-year-old woman presented with left HFS. Magnetic resonance imaging showed a saccular aneurysm of the left VA-PICA which compressed the seventh cranial nerve at its root exit zone (REZ). Stent-assisted coil embolization resulted in intraoperative disappearance of AMR in the intraoperative electrophysiological study and HFS was relieved temporally. One month after endovascular surgery, HFS slightly occurred again with the re-appearance of the AMR, although there was no recurrence of aneurysm. Thereafter, the frequency of her HFS markedly decreased to once per several days 1 year after the coiling.

Conclusion: Although complete disappearance of symptoms was not obtained, it was suggested that coil embolization is one of the therapeutic options for HFS which is caused by aneurysmal compression of REZ and intraoperative AMR is useful for identification of responsible lesions and determination of therapeutic effects.

Keywords ▶ hemifacial spasm, aneurysm, coil embolization, abnormal muscle response

Introduction

Hemifacial spasm (HFS) is caused by compression of root exit zone (REZ) of facial nerve by a cerebral artery in many cases, but, in rare cases, HFS has been reported to be caused by tumor, cerebral arteriovenous malformation, and aneurysm.[1)]

Here, we presented the case of HFS caused by a saccular aneurysm of the vertebral and posterior inferior cerebellar arteries (VA-PICA), which was treated by coil embolization with assistance of abnormal muscle response (AMR) monitoring and showed the marked improvement in symptoms.

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Case Presentation

A 62-year-old woman presented with HFS in her left eyelid, which appeared 3 years ago and progressively aggravated over the last 6 months to be persistent during the daytime. MRI revealed a left VA-PICA aneurysm, which was in the posterior upper direction, located near REZ of the facial nerve. Then we considered the cause of HFS was the aneurysm.

Neurological findings: Her general condition was good and she had no neurological abnormality except for continuous HFS of the left eyelid which did not expand to the orbicularis oris muscle or platysma.

Imaging findings: Computed tomography angiography (CTA) revealed that the left vertebral artery (VA) was tortuous and a saccular aneurysm of the left VA-PICA junction, projecting toward the posterior upper direction (**Fig. 1A–1C**). The aneurysm had a wide neck and its neck slightly rode on the posterior inferior cerebellar artery

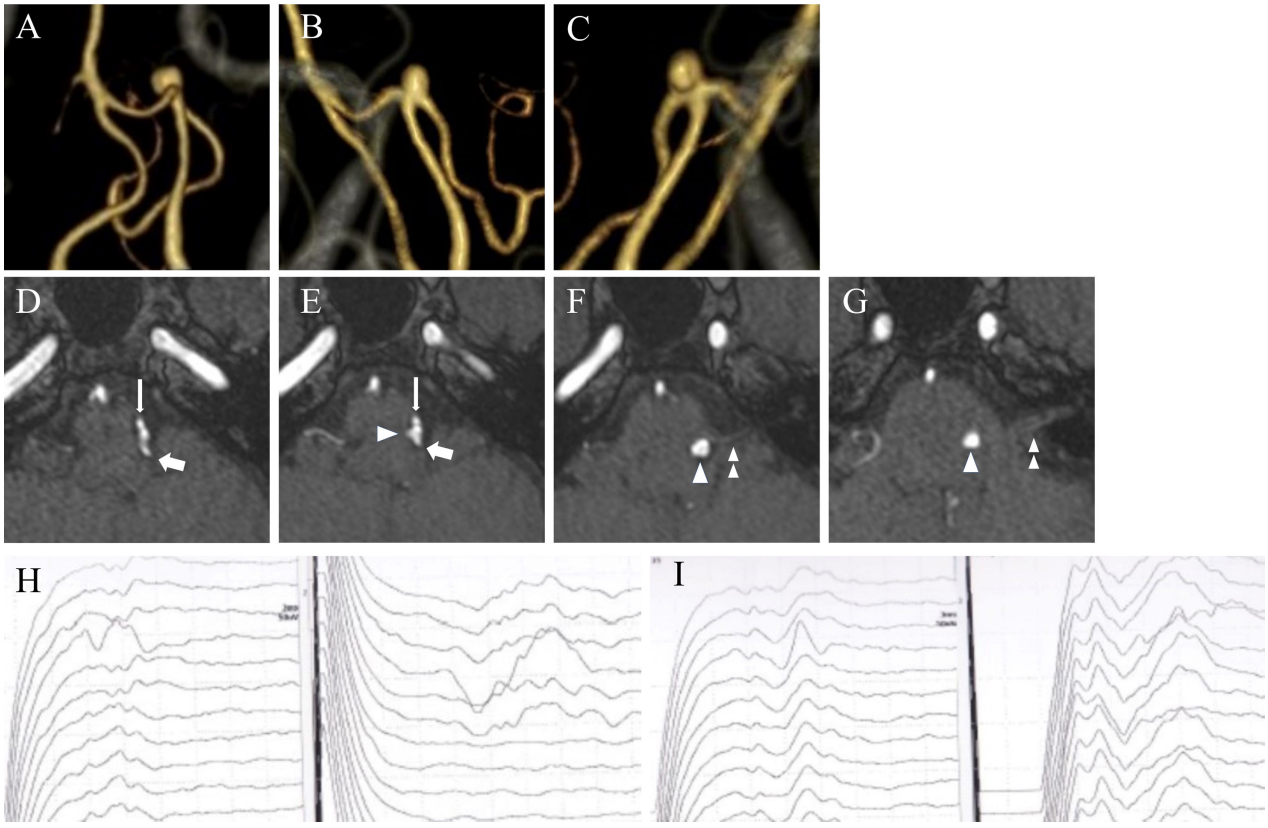

Fig. 1 (**A–C**) Preoperative 3D-CTA images. (**A**) Frontal view, (**B**) lateral view (left side), and (**C**) lateral view (right side). An aneurysm was present at the left VA-posterior inferior cerebellar artery. It was in the posterior upper direction and the bleb headed toward the brainstem (right side). The neck width was 4.2 mm and the dome size was 4.3 mm, showing a wide neck. (**D–G**) Serial sections of original MRA images. The arrow indicates the VA, the large arrow indicates the PICA, the arrow head indicates the aneurysm, and the double arrow head indicates the complex of the seventh and eighth cranial nerves. (**H** and **I**) Preoperative records of facial nerve stimulation-evoked potential. (**H**) Stimulation of the zygomatic branch of the facial nerve. The orbicularis oculi muscle (left) and mentalis muscle (right). AMR was observed. (**I**) Stimulation of the marginal mandibular branch of the facial nerve. The orbicularis oculi muscle (left) and mentalis muscle (right). AMR was observed. 3D-CTA: three-dimensional computed tomography angiography; AMR: abnormal muscle response; MRA: magnetic resonance angiography; PICA: posterior inferior cerebellar artery; VA: vertebral artery

(PICA); the neck width was 4.2 mm, the dome size was 4.3 mm, and the Dome/Neck Ratio was 1.0. Magnetic resonance angiography (MRA) revealed that a dome of the aneurysm was buried in the brain stem and compressing REZ of the left facial nerve. Other blood vessels offending REZ was not clear on MRI and CTA (**Fig. 1D–1G**).

Clinical course: Of course, we first recommend microvascular decompression (MVD) surgery via craniotomy to the patient to treat aneurysm and HFS. But we also explained as follows: the risk of surgical complications was not low because of the location and the projection of the aneurysm as shown in **Fig. 1F** and **1G**; the coil embolization might be safer, although its effect on HFS was unclear; injection of botulinum toxin was considered for alternative treatment when coil embolization was not effective to HFS. The patient finally selected coil embolization.

Then, we planned to perform coil embolization under general anesthesia with the assistance of stent placement on the PICA side because the aneurysm had a wide neck and slightly rode on the PICA. In addition, we also planned to use the AMR monitoring to confirm the effect of the each endovascular procedures on HFS (**Fig. 1H** and **1I**).

In detail, the orbicularis oculi muscle and mentalis muscle were monitored by stimulating the marginal mandibular branch of the facial nerve during surgery, and abnormal electrical potentials by facial nerve stimulation were recorded after microcatheter insertion, stent placement, and intra-aneurysmal embolization, respectively. First, we attempted to navigate a microcatheter from the right VA into the left PICA through the left VA, but this procedure was not completed because of its difficulty. Second, since the neck of the aneurysm was wide, we placed Scepter XC 4×11 mm (Terumo, Tokyo, Japan) from the left VA to the basilar artery for the purpose of balloon-assisted coiling, but this balloon was not considered to appropriate to coiling in this case. At that time, the left VA was straightened

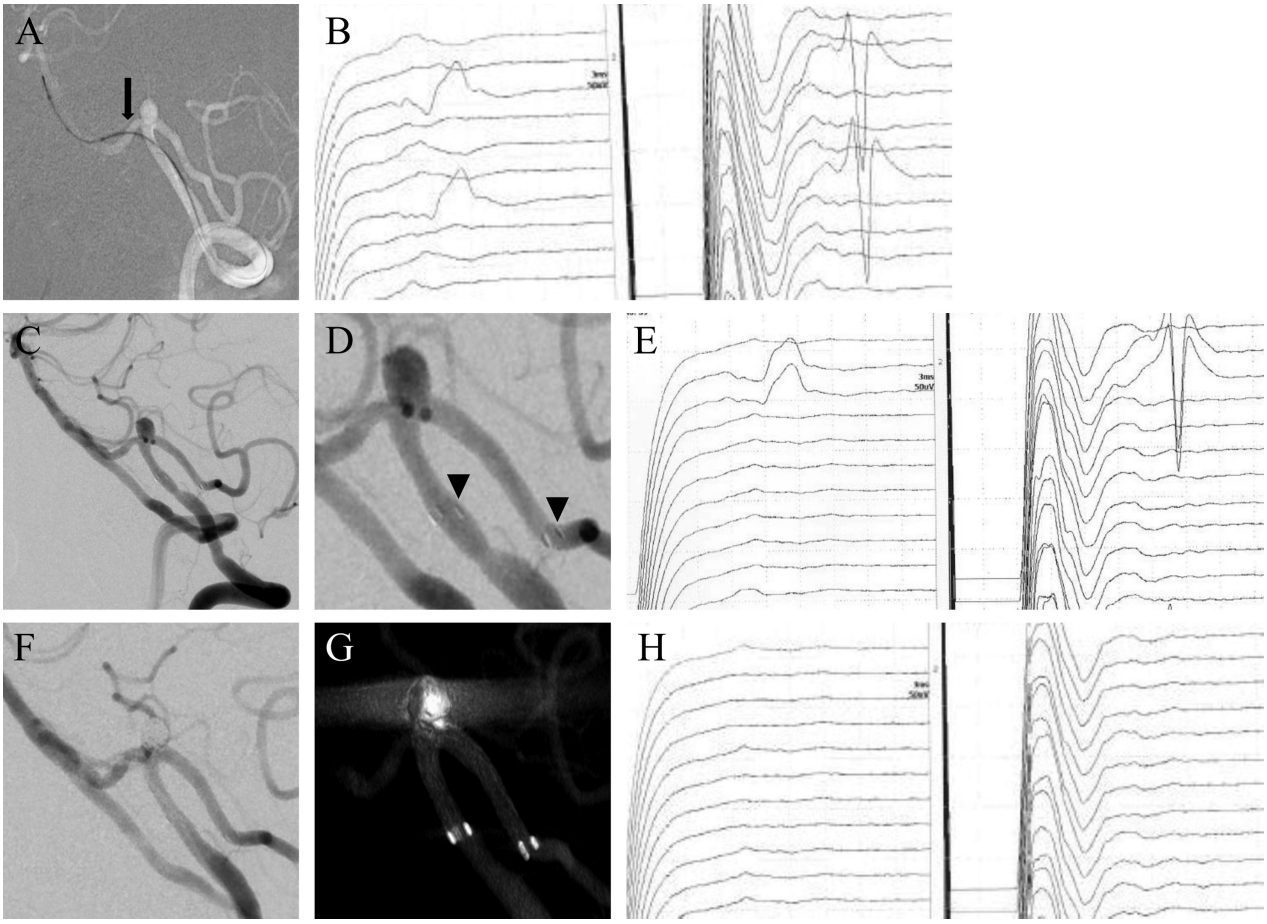

Fig. 2 (**A** and **B**) A microcatheter placed in the left VA over the basilar artery along the road map (**A**). Vascular displacement by catheter insertion was noted (arrow). AMR was noted on evoked potential recording (**B**). (**C–E**) A stent was placed in the posterior inferior cerebellar artery over the VA. Working angle (**C**) and its magnified view (**D**). AMR was noted on evoked potential recording during this procedure. The arrow heads indicate stent markers. (**F–H**) Working angle and 3D images after intra-aneurysmal embolization. AMR disappeared on evoked potential recording during imaging. AMR: abnormal muscle response; VA: vertebral artery

by Scepter, but it did not change AMR at all (**Fig. 2A** and **2B**). Third, Neuroform Atlas 4 × 21 mm (Stryker Japan K.K., Tokyo, Japan) was deployed from the left VA into the left PICA for the assistance of coiling, which procedure also did not change AMR (**Fig. 2C–2E**). Finally, the aneurysm was completely obliterated with coil embolization. In detail, we placed an Axium 3D 3.5 mm × 6 cm coil (Medtronic, Minneapolis, MN, USA) as the first coil. Then, seven additional Axium Prime 3D coils (Medtronic, Minneapolis, MN, USA) were placed; one 3 mm × 6 cm coil, two 2.5 mm × 4 cm coils, three 2 mm × 4 cm coils, and one 1.5 mm × 3 cm coil (**Fig. 2F** and **2G** and **Fig. 3A** and **3B**). Immediately after coil embolization of the aneurysm, AMR completely disappeared (**Fig. 2H**).

Postoperatively, the patient demonstrated no neurological abnormality. The frequency of the patient's facial spasm markedly decreased on the day after the surgery and it was slightly provoked only in the lower eyelid muscles on a blink test. The patient was discharged 10 days after surgery and her HFS completely disappeared at that time. However, spasm of the left lower eyelid muscles slightly recurred 1 month after treatment. The range of facial spasm was narrower and its frequency was lower than that before. As for electrophysiological testing, AMR to stimulation of the facial nerve zygomatic branch remained absent (**Fig. 3E**), but AMR to stimulation of the marginal mandibular branch of the facial nerve re-appeared (**Fig. 3F**). No clear finding of aneurysm recurrence was noted (**Fig. 3C** and **3D**). After the slight recurrence of HFS, her symptoms fortunately started to improve again. One year after the treatment, the frequency of HFS decreased to once per several days.

Discussion

HFS caused by cerebral aneurysms is reported to be rare.[1,2] For treatment of this type of HFS, clipping, endovascular

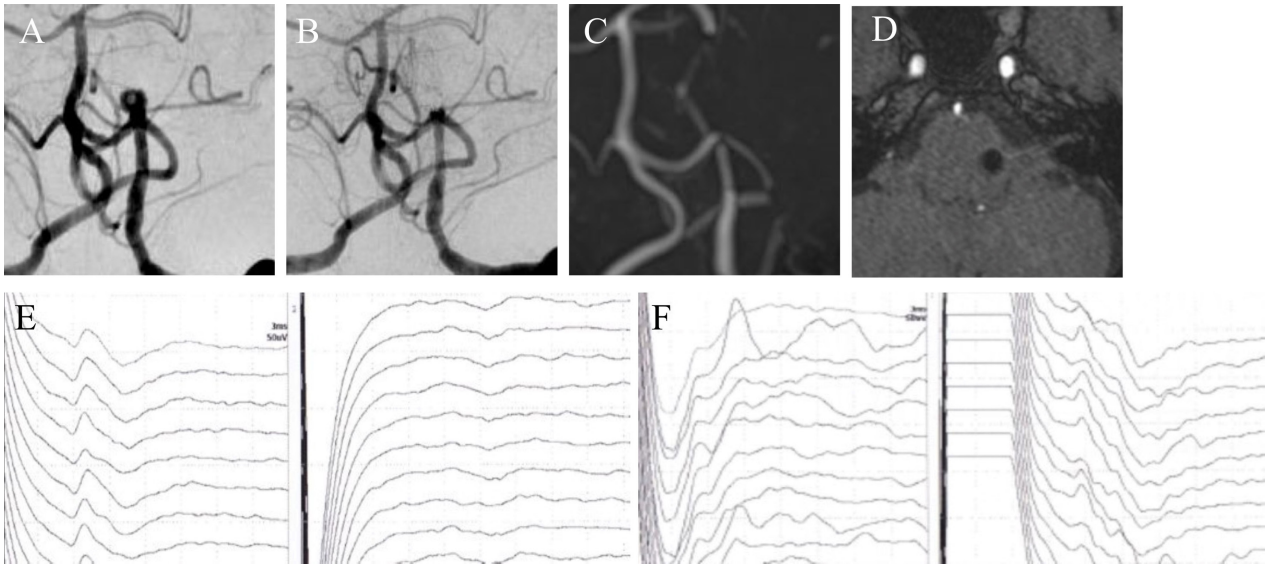

Fig. 3 (**A**) DSA image before embolization, (**B**) DSA image immediately after embolization (**C** and **D**), MRA MIP image 1 month after treatment (**C**) and the original image and MIP image (**D**). No blood inflow into the aneurysm was noted. (**E** and **F**) Facial nerve stimulation-evoked potential recording 1 month after treatment. (**E**) Stimulation of the zygomatic branch of the facial nerve. The orbicularis oculi muscle (left) and mentalis muscle (right). AMR disappeared. (**F**) Stimulation of the marginal mandibular branch of the facial nerve. The orbicularis oculi muscle (left) and mentalis muscle (right). AMR was observed. AMR: abnormal muscle response; DSA: digital subtraction angiography; MRA: magnetic resonance angiography

treatment, and MVD have been reported.[3–8] However, many of these aneurysms were dissecting or fusiform aneurysms. Then, most of the endovascular treatments were parent artery occlusion. As for the saccular aneurysms, there was one reported case in which stent-assisted coil embolization was performed but the patient's HFS did not improved.[9]

HFS is generally treated with MVD via craniotomy. Even in cases that cerebral aneurysm caused HFS, the reliable decompression of REZ can be achieved by MVD because of its visibility and the maneuverability of the offending vessel or aneurysm. However, in our case, the aneurysm was located medial to the cranial nerves VII to XI, and its dome seemed to be buried into the brainstem on MRI, suggesting that the risk of complication of cranial nerve palsy caused by MVD is not low. In fact, lower cranial nerve palsy, including both transient and permanent one, was reported to occur frequently after the clipping of VA-PICA aneurysm (48%).[10] Of course, it was concerned that, when endovascular surgery of an aneurysm did not improve HFS, the subsequent MVD via craniotomy would be difficult because the use of antiplatelet drugs was needed during the perioperative period and the adhesion and the changes in vascular hardness after intravascular coil and stent placement was expected.

Our patient comprehensively judged the risk of surgical procedure and selected endovascular treatment, prioritizing treatment of the aneurysm itself. As for the risk of rupture of the aneurysm which caused HFS, there were a few reports suggesting its risk.[4,11] Moriuchi et al. reported a case in which HFS exacerbated aggressively because of aneurysmal compression of REZ and the very thin aneurysmal wall was observed intraoperatively. Therefore, they proposed that rupture of the aneurysm was threatened.[4] Yuan et al.[11] reported a patient presenting with HFS caused by fusiform aneurysm, who was not treated because of the potential complication of surgery, and died of subarachnoid hemorrhage 2 months after discharge. Of course, no definite conclusion can be drawn based on these case reports alone, but it might be reasonable not to consider that the risk of rupture is low when the aneurysm is symptomatic.

Regarding the mechanism of HFS of this patient, intraoperative AMR monitoring was performed to identify whether it was due to the direct compression by the aneurysm or the compression by other vessels. AMR monitoring has recently been applied as intraoperative monitoring in microvascular surgery for HFS, stimulating the temporal branch of the facial nerve and recording evoked potential (latency: about 10 ms) from the orbicularis oculi muscle or mentalis muscle.[12] Yamashita et al.[13] reported one HFS case in which superselective angiography was performed under local anesthesia to identify the offending vessel and the patient's HFS actually disappeared when they inserted a microcatheter into

the target blood vessel and straightened it. In addition, Murakami et al. reported a case in which AMR was recorded during endovascular parent artery occlusion in the case of a saccular VA aneurysm manifesting as HFS. They reported that the disappearance of AMR after the VA occlusion was observed and the patient's HFS also disappeared.[5] Therefore, we expected that similar changes in AMR could be observed in response to any of the procedures: insertion of catheter, placement of stenting, or coiling. We also expected that the change of AMR might reveal the etiology of the patient's HFS. In fact, the intraoperative AMR change clarified that the disappearance of arterial pulsation to REZ by coil embolization of the aneurysm was needed to treat HFS in our case. Then, monitoring of AMR for endovascular surgery was suggested to be useful for confirming the decompression of REZ during endovascular surgery as well as during craniotomy surgery. As for the effectiveness of the therapy, it was also suggested that coil embolization alone might be insufficient for treatment of aneurysm-induced facial spasm. It is true our patient's HFS recurred 1 month after surgery and AMR also re-appeared, but the symptoms markedly improved thereafter over a year. In this study, we suggest the potential effectiveness of the combination of endovascular therapy and AMR monitoring in treating HFS patients caused by cerebral aneurysms. However, we experienced only one case and further investigation is needed about the relationship between intraoperative AMR findings and surgical outcome.

Conclusion

We reported a patient with cerebral aneurysm manifesting as facial spasm and treated it with intra-aneurysmal coil embolization with assistance of intraoperative AMR monitoring.

Although complete resolution of the symptoms could not be achieved, the symptoms markedly improved. We suggested that coil embolization might be effective for the HFS to some extent and that intraoperative AMR might be useful to identify the responsible lesion and judge the therapeutic effect.

Disclosure Statement

The authors declare no conflicts of interest associated with this manuscript.

References

1) Nagata S, Matsushima T, Fujii K, et al: Hemifacial spasm due to tumor, aneurysm, or arteriovenous malformation. *Surg Neurol* 1992; 38: 204–209.
2) Li T, Lv X, Wu Z: Endovascular treatment of hemifacial spasm associated with a petrosal DAVF using transarterial Onyx embolization. A case report. *Interv Neuroradiol* 2012; 18: 69–73.
3) Tsuchiya D, Kayama T, Saito S, et al: Hemifacial spasm due to a compression of the facial nerve by a fusiform aneurysm of the vertebral artery: case report. *No To Shinkei* 2000; 52: 517–521. (in Japanese)
4) Moriuchi S, Nakagawa H, Yamada M, et al: Hemifacial spasm due to compression of the facial nerve by vertebral artery-posterior inferior cerebellar artery aneurysm and elongated vertebral artery–case report. *Neurol Med Chir (Tokyo)* 1996; 36: 884–887.
5) Murakami H, Kawaguchi T, Fukuda M, et al: Monitoring of the lateral spread response in the endovascular treatment of a hemifacial spasm caused by an unruptured vertebral artery aneurysm. Case report. *J Neurosurg* 2004; 101: 861–863.
6) Uchino M, Nomoto J, Ohtsuka T, et al: Fusiform aneurysm of the vertebral artery presenting with hemifacial spasm treated by microvascular decompression. *Acta Neurochir (Wien)* 2005; 147: 901–903.
7) Matsumoto K, Saijo T, Kuyama H, et al: Hemifacial spasm caused by a spontaneous dissecting aneurysm of the vertebral artery. Case report. *J Neurosurg* 1991; 74: 650–652.
8) Matsumoto K, Kimura S, Kakita K: Endovascular treatment of vertebral artery aneurysm manifesting as progressive hemifacial spasm. *Neurol Med Chir (Tokyo)* 2005; 45: 360–362.
9) Li C, Li Y, Jiang C, et al: Remission of neurovascular conflicts in the cerebellopontine angle in interventional neuroradiology. *J Neurointerv Surg* 2016; 8: 87–93.
10) Al-khayat H, Al-Khayat H, Beshay J, et al: Vertebral artery-posteroinferior cerebellar artery aneurysms: clinical and lower cranial nerve outcomes in 52 patients. *Neurosurgery* 2005; 56: 2–10; discussion 11.
11) Yuan F, Lin J, Ding L, et al: Hemifacial spasm and recurrent stroke due to vertebrobasilar dolichoectasia coexisting with saccular aneurysm of the basilar artery: a case report. *Turk Neurosurg* 2013; 23: 282–284.
12) Higuchi Y, Hirono S, Horiguchi K et al: Microvascular decompression for hemifacial spasm: pathophysiology and intraoperative monitoring. *Jpn J Neurosurg (Tokyo)* 2017; 26: 263–272. (in Japanese)
13) Yamashita K, Hojo M, Okamoto S, et al: Possible role of neurointerventional techniques in the diagnosis of hemifacial spasm. *AJNR Am J Neuroradiol* 1997; 18: 287–290.