A Case of Vertebral Artery Aneurysm Causing Hemifacial Spasm Rapidly Improved after Parent Artery Occlusion

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Objective: The authors report a rare case of symptomatic unruptured fusiform vertebral artery (VA) aneurysm causing hemifacial spasm, which was successfully treated by endovascular parent artery occlusion (PAO).

Case Presentation: A 56-year-old man presented with left hemifacial spasm, and the symptom progressed rapidly over 3–4 weeks including difficulty of eye opening. Brain MRI showed a left fusiform VA aneurysm with a maximum diameter of approximately 10 mm, which compressed the root exit zone (REZ) of the left facial nerve. Endovascular PAO of the left VA was performed with coils. The hemifacial spasm disappeared immediately after PAO. The size of the aneurysm was markedly reduced on MRI on the next day. No recurrence of the hemifacial spasm and aneurysm was observed after half a year after PAO.

Conclusion: This rare case suggested that endovascular PAO may be an effective treatment for hemifacial spasm caused by a relatively large aneurysm.

Keywords ▶ hemifacial spasm, vertebral artery, aneurysm, endovascular therapy

Introduction

Hemifacial spasm is a symptom of neurovascular compression syndrome and, generally, the responsible artery with aging-associated arteriosclerosis presses the root exit zone (REZ) of the facial nerve near the brain stem and gradually causes the symptom.1 Botulinum toxin treatment and drug therapy are performed as symptomatic treatment, and microvascular decompression (MVD) is performed as curative treatment. The usefulness of MVD is widely recognized also in Japan based on surgical cases reported by Jannetta et al. in the 1970s,2 and it has spread as the standard treatment because the lesion can be safely approached by conventional suboccipital craniotomy.

Hemifacial spasm caused by an aneurysm is rare. Many reported cases were treated with direct surgery, but endovascular-treated cases are rare.

We encountered a rare case of symptomatic unruptured fusiform vertebral artery (VA) aneurysm which caused hemifacial spasm and rapidly progressed. It was treated with endovascular parent artery occlusion (PAO) and the symptom rapidly disappeared immediately after treatment. Endovascular treatment may be a useful therapeutic method for hemifacial spasm caused by an aneurysm. We report the outcome of treatment with a literature review.

Case Presentation

The patient was a 56-year-old male with a chief complaint of left hemifacial spasm. The patient was being treated with oral drugs for hypertension. Left hemifacial spasm appeared from the palpebral part and it rapidly advanced to the buccal muscles, levator angli oris muscle, and platysma. Tonic-clonic convulsion persisted, leading to difficulty of eye opening. Left unruptured fusiform VA aneurysm was found on MRA of the head by a physician.
contralateral VA was mostly the same as that on the affected side, the balloon occlusion test (BOT) of the left VA was not performed. Antiplatelet drug administration of aspirin 100 mg/day and clopidrel 75 mg/day was initiated 1 week before treatment. Systemic heparinization was administered under general anesthesia, and the activated clotting time (ACT) was set at 200–250 seconds. In the system, 6Fr. Optimo (Tokai Medical, Aichi, Japan) was placed in the left VA through the right femoral artery, and then Excel-sior SL-10 (STR; Stryker, Kalamazoo, MI, USA) was guided into the aneurysm using Silverspeed 10 (Medtronic, Minneapolis, MN, USA). The balloon of Optimo was inflated to control proximal flow. Using Target (Stryker) XL 360 soft 9×30 as the first coil, the aneurysm was packed by intentionally winding the coil roughly back to the artery and a region about 1.5 cm in length to the proximal VA was embolized with six target coils and three AXIUM coils (Medtronic), nine coils in total, to occlude the parent artery (Fig. 3A and 3B). The normal artery distal to the aneurysm was not embolized because induction of ischemic complication was of concern. The absence of visualization of the aneurysm was confirmed by angiography of the contralateral VA. Since the aneurysm had a shape easy to access from the contralateral VA, it was decided to perform additional embolization of the aneurysm from the opposite side in case the aneurysm recurred, and treatment was completed (Fig. 3C).

Hemifacial spasm disappeared after awakening from anesthesia. The aneurysm was markedly shrunk on MRI on the day following the treatment (Fig. 4A). After the treatment, argatroban was continuously administered at 60 mg/day for 2 days. No new neurologic manifestation appeared and no abnormality was noted on postoperative diffusion-weighted imaging (DWI). The patient was discharged ambulatory 3 days after treatment at a modified Rankin Scale (mRS) of 0. The antiplatelet drugs were withdrawn one by one and discontinued after 2 months. As of 6 months after treatment, hemifacial spasm did not recur and the aneurysm remained shrunken on MRI (Fig. 5A).

### Discussion

The most frequent cause of hemifacial spasm is vascular compression, and induction of compression by the VA aneurysm is rare, accounting for 0.2%–0.6% of cases. Hemifacial spasm caused by tumor, hemangioma, and cerebral arteriovenous malformation has also been reported. Hemifacial spasm rapidly progressed due to the aneurysm in the...
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reported,\(^3\) and if direct surgery had been selected for the present patient, it would have been difficult because surgery is to remove compression of the nerve by a relatively large aneurysm. Thus, endovascular treatment was selected. However, to our knowledge, there are four reported cases of endovascular treatment of hemifacial spasm. All cases were reported from Japan and treated with PAO.\(^3,5,12\) PAO including tight packing of the aneurysm,\(^4,12\) PAO on the proximal side alone,\(^3\) and endovascular trapping without embolizing aneurysm\(^5\) were performed, and endovascular treatment was effective in all reported cases. In our patient, hemifacial spasm disappeared immediately after awakening from anesthesia. Regarding improvement of hemifacial spasm, symptoms disappeared 3–6 months after treatment.

The present case was treated aiming at treating of facial spasm and prevention of expansion of the aneurysm. Regarding direct surgery, treatment with MVD alone and addition of clipping, wrapping, trapping, and occlusion of the proximal region have been reported.\(^6,10,11\) and symptoms disappeared or alleviated immediately after treatment. However, hearing impairment, facial palsy, and cerebellar infarction caused by general MVD have been reported.\(^13\) and if direct surgery had been selected for the present patient, it would have been difficult because surgery is to remove compression of the nerve by a relatively large aneurysm. Thus, endovascular treatment was selected. However, to our knowledge, there are four reported cases of endovascular treatment of hemifacial spasm. All cases were reported from Japan and treated with PAO.\(^3,5,12\) PAO including tight packing of the aneurysm,\(^4,12\) PAO on the proximal side alone,\(^3\) and endovascular trapping without embolizing aneurysm\(^5\) were performed, and endovascular treatment was effective in all reported cases. In our patient, hemifacial spasm disappeared immediately after awakening from anesthesia. Regarding improvement of hemifacial spasm, symptoms disappeared 3–6 months after treatment.
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in all reported cases treated with PAO. The reason for disappearance of the symptoms immediately after treatment was unclear, but the aneurysm was packed intentionally roughly to prevent the mass effect and this may have prevented unnecessary compression of REZ by the coil. Nagashima et al.\(^3\) also reported that slight change in the vascular position and hemodynamic changes may have improved symptoms. When direct surgery is performed, symptoms disappear or remit immediately after surgery because compression of REZ is physically removed, whereas endovascular treatment may require time to reduce the mass effect although obstruction of the parent artery reduces vascular pulsation and pressure in the aneurysm. Even though vascular pulsation was weakened by PAO, thrombosed aneurysm and coils may have acted as a mass effect and prevented rapid decompression of REZ. It is controversial whether the aneurysm should be tightly packed. Nakagawa et al.\(^12\) reported PAO including embolization of an aneurysm. They stated that simple PAO is insufficient to inhibit pulsation of an aneurysm due to reflux from the contralateral VA, and complete blockade of blood flow into the aneurysm and inhibition of pulsation of facial nerve REZ through the artery by embolization of both the aneurysm and parent artery leads to improvement of symptoms.

Fig. 4 (A) Marked shrinkage of the aneurysm was noted on postoperative T2-weighted MRI. (B) The left VA aneurysm was not visualized on postoperative MRA. VA: vertebral artery

Fig. 5 (A) The aneurysm remained shrunken on MRI 6 months after treatment. (B) No recurrence of aneurysm was noted on MRA 6 months after treatment.
Therefore, although it should be investigated in each case, endovascular PAO is a useful treatment option. Stent-assisted embolization of aneurysm aiming at conservation of the parent artery should also be investigated, but when the aneurysm is spindle-shaped, tight packing of the space around the stent is necessary, for which reduction of the mass effect cannot be expected. In addition, the possibility of resection of the aneurysm by direct surgery in case of no remission of symptoms by PAO was considered. In such cases, when a stent is placed in the VA, resection of the aneurysm may be difficult. PAO was selected because of this reason. To our knowledge, treatment of aneurysm manifesting as hemifacial spasm with stent-assisted coil embolization or using a flow diverter has not been reported. At present, it is unclear whether stent-assisted coil embolization remits aneurysm pulsation and mass effect and improves symptoms. When PAO is difficult due to the PICA branching from aneurysm, the indication of endovascular treatment has to be carefully investigated and stent-assisted coil embolization is a treatment option.

Conclusion

We encountered a patient with a rare symptomatic unruptured fusiform VA aneurysm which responded to endovascular treatment. Although the standard treatment for hemifacial spasm is MVD, it was suggested that endovascular PAO is also effective radical treatment of hemifacial spasm considered to be caused by a relatively large aneurysm.

Disclosure Statement

None of the first and co-authors have conflict of interest.

References