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Case Report

Microvascular decompression for hypoglossal nerve palsy secondary to vertebral artery compression: A case report and review of the literature

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Abstract

Background: Neurovascular-compression syndrome (NCS) is described as a prominent pathological contact between cranial nerves and vessels. Trigeminal neuralgia, hemifacial spasm, and glossopharyngeal neuralgia are typical clinical entities associated with NCS. On the other hand, the hyoglossal nerve is rarely affected by NCS.

Case Description: We present a case of hypoglossal nerve palsy (HNP) secondary to vertebral artery (VA) compression. A 47-year-old man presented to our hospital with a 1-month history of dysarthria and dysphagia. Neurological examination revealed left HNP, with an intact swallowing reflex and no oropharyngeal or palatal weakness. Magnetic resonance imaging (constructive interference in steady state) revealed left hypoglossal nerve compression by the V4 segment of the left atherosclerotic VA. He underwent microvascular decompression (MVD) surgery. Intraoperatively, the VA was compressing the hypoglossal nerve. The left VA was moved and attached to the dura matter using a polytetrafluoroethylene (Teflon®) sheet and fibrin glue. Postoperatively, the patient exhibited gradual recovery of HNP in 3 months without dysfunction of lower cranial nerves.

Conclusion: In patients with isolated HNP, vascular compression should be considered as a cause of these symptoms, and subsequent MVD can lead to resolution.

Key Words: Hypoglossal nerve palsy, microvascular decompression, vertebral artery

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INTRODUCTION

Neurovascular compression syndromes (NCS) occur when cranial nerves are compressed by intracranial vascular loops. Trigeminal neuralgia, hemifacial spasm, and glossopharyngeal neuralgia are typical clinical entities associated with NCS. These diseases usually occur when the root exit zone of the nerves is affected. Microvascular decompression (MVD) surgery is effective and usually results in immediate postoperative improvement of symptoms. The hyoglossal nerve is rarely affected

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by NCS, but we encountered a case of hypoglossal nerve palsy (HNP) secondary to vertebral artery (VA) compression.

CASE REPORT

History and examination

A 47-year-old man presented to our hospital with a 1-month history of dysarthria and dysphagia. He did not show swallowing difficulty, but had subtle difficulty in moving his tongue so that he frequently unintendedly bit his tongue. He had a past history of multiple cerebral aneurysms and had a checkup by angiography 6-months prior to the onset. He consumed alcohol occasionally, and had never smoked cigarettes. Neurological examination revealed isolated left HNP [Figure 1a].

Imaging studies

Magnetic resonance imaging (MRI) (CISS, constructive interference in steady state) revealed left hypoglossal nerve compression by the V4 segment of the left atherosclerotic VA [Figure 2]. Further investigation revealed that his intracranial arteries were partially dilated and tortuous as a result of atherosclerotic changes. Multiple aneurysms were revealed including a partially thrombosed large left MCA saccular aneurysm and a fusiform aneurysm at the left PICA. The left VA was also dilated and tortuous. The hypoglossal nerve was composed of two bundles, and both of them were running along the left VA in a tortuous pathway to the hypoglossal canal. In contrast, the contralateral hypoglossal nerve was seen linearly and very clearly on the same images. This finding strongly suggested that the hypoglossal nerve was compressed by the left VA [Figure 3].

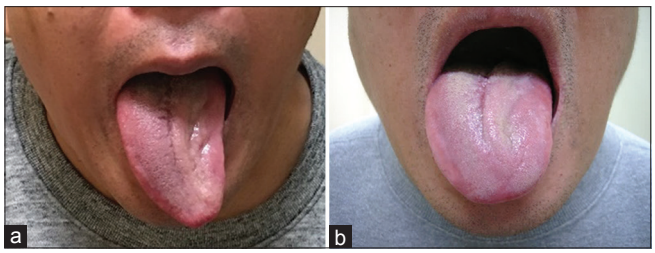

Figure 1: A 47-year-old man with a one-month history of dysarthria showed isolated left hypoglossal nerve palsy (a), which was improved significantly three months after microvascular decompression (b)

Surgical procedure

On the basis of the above diagnosis, he underwent MVD of the left hypoglossal nerve. In a park-bench position, retrosigmoid lateral suboccipital craniotomy enabled us to visualize the glossopharyngeal, vagal, accessory, and hypoglossal nerve together with the VA. Intraoperatively, the VA was compressing the hypoglossal nerve. The hypoglossal nerve was stretched and was extremely thin. The left VA was moved toward the deep side of the surgical field and attached to the dura matter with a polytetrafluoroethylene (Teflon®) sheet and fibrin glue. The left hypoglossal nerve became free from compression after the procedure [Figure 4a and 4b].

Postoperative course

He was discharged 7 days after the operation without any obvious complication. At a 3-month postoperative follow-up, he exhibited improvement of hypoglossal nerve palsy without lower cranial nerve dysfunction [Figure 1b].

DISCUSSION

Stino *et al.*[11] and Keane *et al.*[4] reviewed cases of isolated HNP. According to their reports, possible causes of HNP include tumor, trauma, stroke, hysteria, surgery, multiple sclerosis, infection, Guillian-Barre neuropathy, and radiation. HNP in almost one-half of the patients in Keane's investigation was caused by neoplasia, and over one-half of those were malignant. The next most common cause was trauma. The majority of the palsies were associated with other cranial nerves. There were no patients in their reports who showed isolated HNP due to VA compression. In our case, we believe the atherosclerotic VA compressed the hypoglossal nerve and finally caused HNP. To date, there have been only 12 reported cases of HNP secondary to VA compression, including the present case. A summary of these cases is shown in Table 1.

This pathophysiology was first described in English by Rollnik *et al.*[9] as hypoglossal-vertebral entrapment syndrome. Morini *et al.*[8] reported two cases of isolated HNP due to VA compression. In their cases, both of the patients were treated conservatively and did not show any improvement of their symptoms.

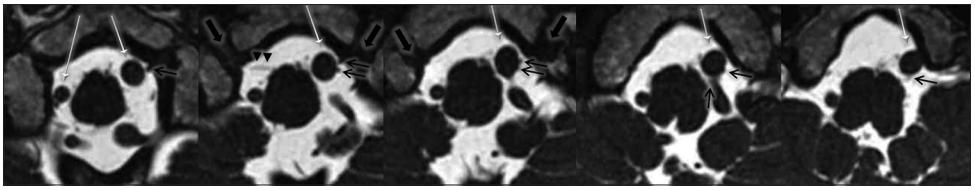

Figure 2: MRI (CISS, constructive interference in steady state) images of the present case. Left hypoglossal nerves (black thin arrows) originated from the ventral side of the medulla oblongata, which is composed of two bundles, and ran into the hypoglossal canal (black thick arrows) beyond the left VA (white thin arrow). It is easy to detect when compared with the contralateral side (arrow head)

Table 1: Twelve cases of HN palsy due to VA compression

Authors and Year	Age (y.o.) Sex	Symptoms other than tongue deviation	Onset and length of the symptoms	Past history	Compressing vessel	Treatment	Postoperative Course
Rollnik[9] 1996	59 Male	Sialorrhea	5 weeks before admission	ND	Atherosclerotic VA	Conservation	NA
Morini[8] 1998	77 Male	Dysarthria, mild hemiparesis	Transient symptoms only	Odontoid process fx palpebral carcinoma	Atherosclerotic VA	Conservation	Aggravated 22 months later
Morini[8] 1998	48 Male	Tongue deviation only	Noticed 6 months before the admission	ND	Atherosclerotic VA	Conservation	ND
Salvi[10] 1999	64 Female	Tongue deviation only	Noticed 3 years before diagnosis	None	Atherosclerotic VA	Conservation	No change over 7 years
McKeon[6] 2007	54 Male	Tongue weakness	Preceded by a week-long history of severe neck pain	None	Dissected VA	Antithrombotic therapy	Resolved in 2 weeks
Graham[3] 2007	77 Male	Tongue deviation only	Sudden onset and lasted for 3 months	Lymphoma bladder carcinoma	Atherosclerotic VA	Conservation	No change
Aladdin[1] 2008	54 Male	Dysarthria	ND	ND	Atherosclerotic VA	Conservation	ND
Straube[12] 2008	60 Male	Strict unilateral recurrent headache	Gradual onset 7 weeks before diagnosis	Hypertension	VA attached to HN	Conservation	No change
Cheong[2] 2011	32 Male	Dysarthria, dysphagia	Acute aggravation after 1-month dysphagia	None	Normal VA	MVD	Improvement in 3 months
Yamamoto[13] 2011	80 Male	Dysarthria, dysphagia	Gradual onset and progressed over 1 year	ND	Atherosclerotic VA	Conservation	ND
Mahadevappa[5] 2012	20 Female	Neck pain, dysarthria	Sudden onset dysarthria with a preceding 2-week history of neck pain	None	Dissected VA	Antithrombotic therapy	Resolved in 1 month
Present case	47 Male	Dysarthria	Gradual onset and progressed over 1 month	Unruptured MCA and VA aneurysms	Atherosclerotic VA	MVD	Improvement in 3 months

fx: Fracture, VA: Vertebral Artery, ND: Not Described, NA: Not Available, HN: Hypoglossal Nerve, MVD: Microvascular Decompression, MCA: Middle Cerebral Artery

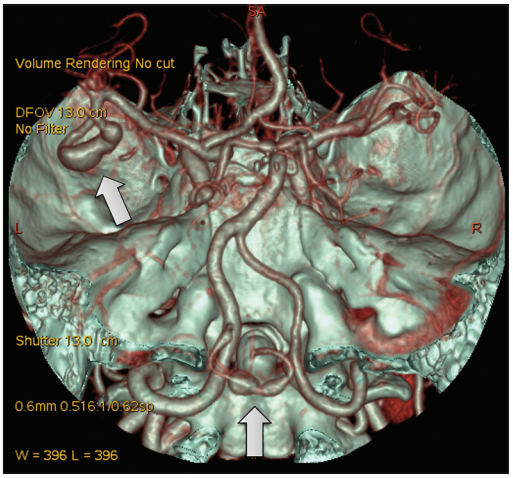

Figure 3: Multiple aneurysms including a partially thrombosed large left MCA saccular aneurysm and a fusiform aneurysm at the left PICA were revealed. The left VA was also dilated and winding

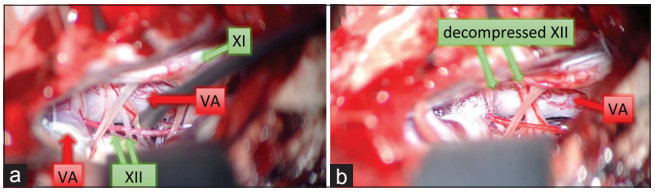

Figure 4: The left vertebral artery compressed the hypoglossal nerve from the deep side of the surgical field to the superficial side. Hypoglossal nerves were stretched by the left VA and became extremely thin (a). The left VA was moved anteriorly and attached to dura using a polytetrafluoroethylene (Teflon®) sheet and fibrin glue. Hypoglossal nerves were released (b)

Graham *et al.*[3] also reported a similar case in 2007, and the patient also did not recover after conservative therapy. Aladdin *et al.*[1] argued that no effective treatment was found for this phenomenon. In 2011, Cheong *et al.*[2] presented a 32-year-old man with a 1-month history of dysarthria and difficulty in tongue movement and swallowing. This is the only patient who underwent surgical intervention among previously reported cases. Intraoperatively, VA was compressing the hypoglossal nerve and interposition of the VA and the

hypoglossal nerve was performed using Teflon®. The patient showed improvement of his tongue deviation at 3 months postoperatively. This case strongly suggests that surgical intervention is effective for this pathophysiology.

In reviewing previous MRI findings, most of the patients who had conservative therapy show atherosclerotic or ectatic changes of the VA, which was similar to our case study. Thus, we believe atherosclerotic changes to the VA caused hypoglossal nerve compression, finally resulting in HNP in these patients. The fact that their symptoms appeared gradually supports the theory that slowly ongoing atherosclerotic changes caused hypoglossal nerve compression. In those patients, prognosis is good when treated with MVD surgery. Two cases of MVD surgery, including ours, have shown good recovery of tongue deviation after 3 months.[2] On the contrary, in other patients who were treated conservatively, the symptoms were unchanged or aggravated. In patients with a preoperative diagnosis of HNP with VA compression, decompression via MVD surgery is strongly recommended.

We also found other interesting case reports with similar pathophysiology. Mahadevappa *et al*.[5] presented a 20-year-old woman with sudden onset dysarthria caused by HNP preceded by a 2-week history of neck pain. MR angiography revealed an ipsilateral dissected VA, which was responsible for the symptom. She was treated with aspirin followed by warfarin. At a 1-month follow-up her tongue deviation had disappeared. Mckeon *et al*.[6] also reported a 54-year-old man with HNP without any other cranial nerve palsies. He was also reported to have undergone VA dissection, and subsequent warfarin intake resolved his symptoms in 2 weeks. These cases indicate the necessity of careful consideration of VA dissection as a differential diagnosis for patients with isolated HNP. For patients with dissected VAs, a conservative therapy with anti-coagulative drugs is recommended. For diagnosis, we should consider the preceding history of sudden onset headaches or neck pain. MRA, which is essential for diagnosis of HNP due to VA compression, is also useful for screening VA dissection as intimal flap or diameter changes, including occlusion, can be detected.[5] Several cases of HNP caused internal carotid artery dissection have also been reported.[7] According to Mokri *et al*.,[7] 5.2% of cervical ICA dissection patients showed lower cranial nerve palsy. In general, conservative treatment is the first choice.

CONCLUSION

To our knowledge, NCS of the hypoglossal nerve is a rare condition. In patients with isolated HNP, vascular compression should be considered as a cause of the symptoms, and subsequent MVD can lead to resolution of the symptoms. We should carefully differentiate HNP due to VA dissection, which can cause similar symptoms, but does not require surgical intervention. As far as we know, this is the first case report with a thorough literature review of hypoglossal nerve palsy by VA compression.

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Conflicts of interest
There are no conflicts of interest.

REFERENCES

1. Aladdin Y, Siddiqi ZA, Khan K. Hypoglossal-vertebral entrapment syndrome. Neurology 2008;71:461.
2. Cheong JH, Kim JM, Yang MS, Kim CH. Resolution of isolated unilateral hypoglossal nerve palsy following microvascular decompression of the intracranial vertebral artery. J Korean Neurosurg 2011;49:167-70.
3. Graham RM, Thomson EF, Baldwin AJ. Isolated hypoglossal nerve palsy due to a vascular anomaly, Int J Oral Maxillofac Surg 2007;36:759-61.
4. Keane JR. Twelfth-nerve palsy. Analysis of 100 cases. Arch Neurol 1996;53:561-6.
5. Mahadevappa K, Chacko T, Nair AK. Isolated unilateral hypoglossal nerve palsy due to vertebral artery dissection. Clin Med Res 2012;10:127-30.
6. Mckeon A, Murphy S, McNamara B, Ryder DQ, Galvin RJ. Isolated hypoglossal nerve palsy due to compression by a dissecting vertebral artery, Eur Neurol 2005;53:162-4.
7. Mokri B, Silbert PL, Schievink WI. Cranial nerve palsy in spontaneous dissection of the extracranial internal carotid artery. Neurology 1996;46:356-9.
8. Morini A, Rozza L, Manera V, Buganza M, Tranquillini E, Orrico D. Isolated hypoglossal nerve palsy due to an anomalous vertebral artery course: Report of two cases. Ital J Neurol Sci 1998;19:379-82.
9. Rollnik JD, Sindern E, Mosler F, Spring B, Malin JP. Isolated peripheral hypoglossal palsy caused by a kinking of the left vertebral artery (hypoglossal vertebral entrapment syndrome). Eur Neurol 1996;36:324-5.
10. Salvi F, Mascalchi M, Plasmati R, Tugnoli V, De Grandis D. Hypoglossal vertebral entrapment syndrome. Muscle Nerve 1999;22:288-9.
11. Stino AM, Smith BE, Temkit M, Nagi Reddy S. Hypoglossal nerve palsy: 245 cases. Muscle Nerve 2016;54:1050-4.
12. Straube A, Linn J. Unilateral headache attacks and ipsilateral atrophy of the tongue due to neurovascular compression of the hypoglossal nerve. Cephalagia 2008;28:996-8.
13. Yamamoto M, Suzuki K, Takekawa H, Hirata K. Isolated hypoglossal nerve palsy caused by neurovascular compression. Intern Med 2001;50:2701-2.