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Case Report

Unilateral isolated hypoglossal nerve palsy due to pathologically adherent PICA fusiform aneurysm – A case report

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Abstract

Background: Isolated hypoglossal nerve palsy due to mechanical compression by a vascular lesion is rare.

Case Description: We report the case of a 72-year-old man who presented with a 4-year history of swallowing disturbance and subsequently progressively worsening left-sided tongue atrophy. He was referred to our department by a neurologist due a magnetic resonance imaging detected left vertebral artery compression of the medulla. Neurological examination was unremarkable except for left hypoglossal nerve dysfunction, which presented as left-sided atrophy and impaired movement of the tongue. Three-dimensional computed tomography angiography showed proximal left posterior inferior cerebellar artery (PICA) origin fusiform aneurysm. Microvascular decompression was done through a left transcondylar fossa approach. Intraoperative findings were thickened arachnoid around the lower cranial nerves, fusiform aneurysm of the left PICA at its origin from the left vertebral artery which was severely adherent to and compressing the left hypoglossal nerve rootlets.

Conclusion: The PICA has a very close relationship to the hypoglossal nerve, and its fusiform dilatation could cause isolated hypoglossal nerve dysfunction. Pathological adhesions between hypoglossal rootlets and the PICA aneurysm wall could be a possible contributor in the development and progression of hypoglossal nerve palsy.

Key Words: Adhesion, chronic arachnoiditis, fusiform aneurysm, hypoglossal nerve palsy, vascular compression

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INTRODUCTION

Hypoglossal nerve (HN) palsy is not an uncommon neurological disease. It has varied etiologies and is usually associated with dysfunctionality of other lower cranial nerves.[4] Isolated HN palsy due to compression from vascular pathologies is very rare. It is limited to a few reports and case series on mechanical compression of the HN by the vertebral artery and persistent fetal

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vessels.[3] Enlarged emissary vein in the hypoglossal canal as a cause has also been reported.[12] We present the case of a 72-year-old man with a progressively worsening left HN palsy due to compression of HN rootlets by a pathologically adherent fusiform aneurysm of left posterior inferior cerebellar artery (PICA) origin.

CASE DESCRIPTION

History

A 72-year-old man was referred from a local hospital with a 4-year history of swallowing difficulty which progressively worsened over time. He also had occasional episodes of aspiration. There was no history of neck trauma, headache, dysarthria, or gait problems. He was a known hypertensive but not a diabetic. He had a positive history of subarachnoid hemorrhage (SAH) from a ruptured middle cerebral artery aneurysm, which was managed surgically (not clipped) at a local hospital 35 years before the present illness. Laryngeal endoscopy showed no obstructive lesion, however, videoendoscopy and videofluoroscopy revealed delayed swallowing reflex and occasional aspiration.

In our hospital, he was initially managed by the otorhinolaryngologists (ORL) as an outpatient. He developed left-sided tongue atrophy [Figure 1] which progressively worsened during ORL follow-up. He was later referred to the neurologists who made a diagnosis of left HN palsy caused by magnetic resonance imaging (MRI) [Figures 2a-c] detected left vertebral artery (VA) compression of the medulla. Three-dimensional computed tomography angiography showed left PICA fusiform aneurysm [Figure 2d]. He was then referred to the neurosurgery service for microvascular decompression (MVD).

Examination

General physical examination was unremarkable. Neurological examination showed an alert elderly man with intact higher cerebral function. He had left tongue atrophy and impaired movement of the tongue. Other cranial nerves were intact. Sensorimotor system examination was normal and coordination was good. He was worked up for MVD after an informed consent was obtained.

Operation and postoperative course

The MVD was done through a left transcondylar fossa approach. Intraoperative findings were thickened arachnoid around the lower cranial nerves and fusiform aneurysm of the left PICA at its origin from the left VA which was severely adherent to and compressing the left HN rootlets [Figure 3a]. The HN rootlets were carefully dissected from the aneurysm wall [Figure 3b and c]. The PICA with the aneurysm was transposed caudally and anchored to the dura with three pieces of oxidized cellulose and fibrin glue [Figure 3d]. The PICA aneurysm looked difficult to clip and wrapping was not done for fear

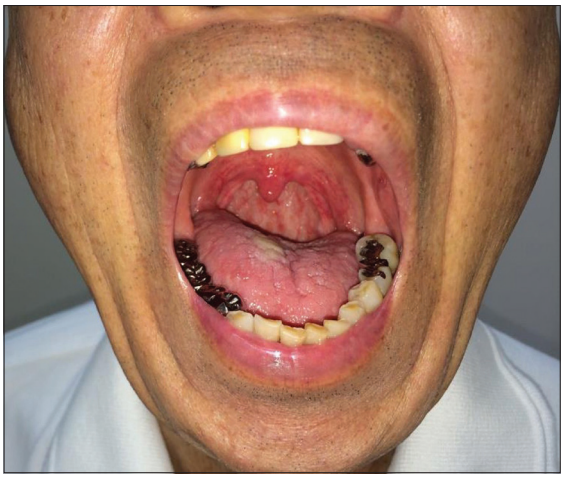

Figure 1: Preoperative picture of the patient showing left-sided tongue atrophy but no deviation

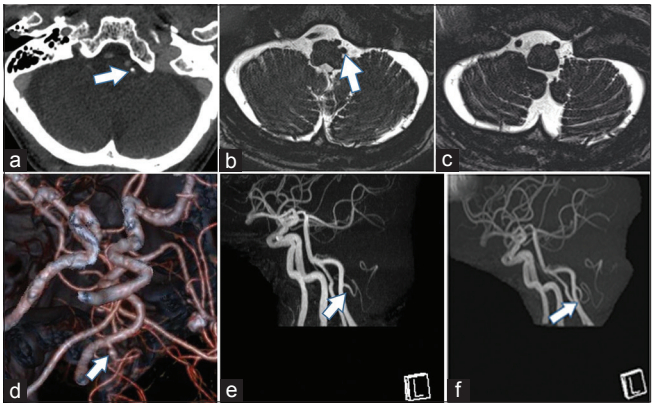

Figure 2: Preoperative images: plain CT (a) showing PICA aneurysm wall calcification (arrow); axial T2-WI (b and c) showing the left VA and PICA (arrow) running very close to the left HN rootlets (arrow); CTA (d) showing left PICA origin fusiform aneurysm (arrow); MRA showing the PICA (arrow) before (e) and after (f) transposition (arrow)

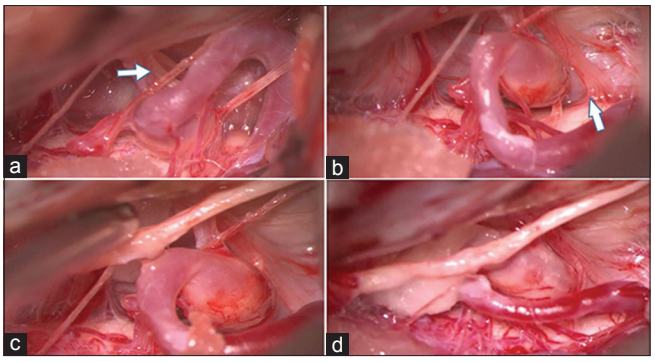

Figure 3: Images captured intraoperatively: showing adhesions between the fusiform PICA aneurysm and the HN rootlets (arrow) (a); successful separation of the aneurysm (arrow) from the HN rootlets (b and c); transposition of the PICA with oxidized cellulose (d)

of inducing further inflammatory response and worsen the adhesions around the already dysfunctional HN. No complications were encountered during the procedure.

Postoperatively, the patient's preoperative neurological status was preserved. MR Angiography showed anterior medullary segment of PICA was successfully transpositioned without stenosis [Figure 2e and f]. There was no objective improvement in his tongue atrophy at 1-year follow-up, however, there was subjective progressive improvement in his tongue movement and swallowing. MRI done at 1-year follow-up was not significantly different from the immediate postoperative MRI. Videoendoscopy and fluoroscopy, however, have remained the same.

DISCUSSION

The HN is a pure motor nerve that innervates all intrinsic and extrinsic muscles of the tongue except the palatoglossus muscle.[3] It has five segments – medullary, cisternal, skull base, nasopharyngeal, and sublingual. The HN (cisternal segment) emerges from the medulla as a series of 3 to 15 rootlets in the preolivary sulcus and exits the posterior fossa through the hypoglossal canal.[10] The HN can be damaged anywhere along its course from the brainstem to the tongue. Different disorders, for the different segments, that can cause HN palsy have been described in literature and these include vascular, inflammatory, traumatic, neoplastic, or degenerative.[5,7] Nuclear lesions tend to give bilateral palsy due to the close proximity of HN nuclei in the brainstem as opposed to peripheral lesions.[6]

The hypoglossal rootlets are closely related to both the VA and PICA. The relationship of the HN rootlets to PICA varies significantly. The PICA can arise below, above, or at the level of the rootlets. Those that arise below or above the rootlets usually course superior or inferior to the rootlets rather than through them. Those that arise at the level of the rootlets usually course in between and frequently stretch the hypoglossal rootlets.[10] Because of this close relationship, vertebrobasilar pathologies such as fusiform PICA origin aneurysm can further stretch and compress the rootlets resulting in nerve palsy.[1,7,9]

There have been a few reports in literature of isolated HN palsy caused by mechanical compression by normal or abnormal intracranial vessels.[3,7,8,11,12] The index case had a past history of SAH, which was surgically managed probably by wrapping. The SAH and possibly inflammation around the PICA fusiform aneurysm might be responsible for the development of the adhesions between the aneurysm and HN rootlets. The rootlets were so severely adherent to aneurysm wall that it was very difficult to separate them. It is possible the adhesions might have contributed to the progression of the HN palsy probably by fixing and restricting movement between the rootlets and the aneurysm. This might worsen the effect of arterial pulsation and mechanical compression from the fusiform aneurysm on the rootlets, which are known mechanisms of cranial nerve dysfunction at the Obersteiner–Redlich zone. The subsequent development of progressively worsening tongue atrophy might be from progressive increase in the size of the aneurysm. We speculate that chronic arachnoiditis around the lower cranial nerves might be responsible for the dysphagia which preceded the tongue atrophy.

Fusiform PICA aneurysms are difficult to treat and direct clipping is usually not possible due to circumferential dilatation of the vessel. Surgical approaches previously described include clip/wrapping, segmental sacrifice, flow reversal with proximal occlusion, distal occlusion, and bypass with trapping.[2] Wrapping might elicit further inflammatory reaction and adhesions around the rootlets and hence it was not done. However, close monitoring of the patient and possible aneurysm trapping at a later date were opted for. Postoperatively, there has been some subjective improvement in swallowing. Objective improvement is expected to be slow but progressive as the nerve gradually recovers from the prolonged insult.

CONCLUSION

The PICA has a very close relationship to the HN and its fusiform dilatation could cause isolated HN dysfunction. Pathological adhesions between the HN rootlets and the aneurysm wall could be a contributor in the progression of nerve dysfunction.

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Conflicts of interest
There are no conflicts of interest.

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