

Successive occurrence of vertebrobasilar dolichoectasia induced trigeminal neuralgia, vestibular paroxysmia and hemifacial spasm

A case report

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Abstract

Rationale: Study reported an extremely rare case of trigeminal neuralgia, vestibular paroxysmia, and hemifacial spasm successively occurring in a patient with vertebrobasilar dolichoectasia (VBD).

Patient concerns: A 66-year-old female patient presented with episodic vertigo for 20 days before she was admitted to our hospital. Vertigo suddenly occurred when the patient rotated her head 20 days ago, the symptoms of vertigo were improved after continuous 1 to 3 seconds, and similar symptoms were repeated in sleep and rest, with a frequency of 30 to 40 times per day. The patient had a history of hypertension with poor blood pressure control for more than 20 years.

Diagnoses: The final diagnosis was vertebrobasilar dolichoectasia, right trigeminal neuralgia, and vestibular paroxysmia.

Interventions: Vitamin B1 (10 mg), methylcobalamin (0.5 mg), and carbamazepine (0.1 g) were given orally 3 times a day to relieve the symptoms.

Outcomes: On the seventh day of drug treatment, the symptoms of paroxysmal vertigo and trigeminal neuralgia were completely relieved, but occasional episodes occurred during the follow-up period. Five months after discharge, right hemifacial spasm appeared in the patient, which did not affect the quality of life of the patient, so the patient did not choose further treatment. Six months after discharge, the patient was lost to follow-up.

Lessons: Comprehensive treatment to control VBD risk factors, delay the progression of VBD, and improve clinical symptoms may have a better effect on such patients. However, further research is needed.

Abbreviations: 3D-CISS = 3-dimensional constructive inference in steady state, 3D-TOF-MRA = 3-dimension time-of-flight magnetic resonance imaging, CN = cranial nerve, CTA = computed tomography angiography, GN = glossopharyngeal neuralgia, HFS = hemifacial spasm, MRI = magnetic resonance imaging, NVCS = neurovascular compression syndrome, TN = trigeminal neuralgia, VBD = vertebrobasilar dolichoectasia, VP = vestibular paroxysmia.

Keywords: hemifacial spasm, neurovascular compression syndrome, vertebrobasilar dolichoectasia

1. Introduction

Trigeminal neuralgia, vestibular paroxysmia, and hemifacial spasm all belong to the category of neurovascular compression syndrome (NVCS). At present, most of the reports on these

diseases are associated with indirect compression of the small vascular loops^[1] of the anterior inferior cerebellar artery and superior cerebellar artery located in the branches of vertebrobasilar artery. Direct compression of vertebral basilar artery is relatively not common. In this study, we reported a extremely rare case of trigeminal neuralgia, vestibular paroxysmia, and hemifacial spasm successively occurred in a patient with vertebrobasilar dolichoectasia (VBD).

2. Case report

A 66-year-old female patient presented with episodic vertigo for 20 days before she was admitted to our hospital. Vertigo suddenly occurred when the patient rotated her head 20 days ago, the symptoms of vertigo were improved after continuous 1 to 3 seconds, and similar symptoms were repeated in sleep and rest, with a frequency of 30 to 40 times per day. Occasional tinnitus was found, but the patient had no dysphagia, no obvious weakness of the limbs, and no abnormal sensation. The patient had a 3-year history of trigeminal neuralgia, characterized by right face pain caused by face washing, brushing, or eating, and lasted for a few seconds to several minutes. Oral administration of carbamazepine was discontinuously taken for 1 year, but she stopped carbamazepine on her own for nearly 2 months. The

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patient had a history of hypertension with poor blood pressure control for more than 20 years, the highest blood pressure was 200/100 mm Hg. Physical examination showed poor spirit status, and when touching the right cheek of the patient, it can sometimes cause facial pain, and the common distribution areas of V1 and V2 were affected, cheek, frontal, and parietal regions, upper eyelid, nasal dorsum skin, and other areas were included. There was a decreased sensation in the V1 and V2 regions, and no abnormalities were detected in the rest of the nervous system. The total cholesterol was 6.40 mmol/L, the glucose was 6.65 mmol/L, the potassium was 3.38 mmol/L, and the sodium was 122.3 mmol/L. The routine test of blood, blood clotting functions, and urine were normal. Dix-Hallpike test was negative. The head magnetic resonance imaging (MRI) showed small lacunar infarctions on both sides of the basal ganglia. The head computed tomography angiography (CTA) revealed tortuosity and reflection of the proximal segment of the C1 segment of the left internal carotid artery, thus VBD was confirmed. Three-dimensional constructive inference in steady state (3D-CISS) and 3-dimension time-of-flight MRI (3D-TOF-MRA) (Fig. 1) showed that the right vertebral artery compressed the right vestibulocochlear nerve, and the dilated basilar artery compressed the right trigeminal nerve; this caused the right trigeminal nerve to move upward and outward. Bilateral vertebral arteries were elongated, but the extension of the right vertebral artery and the dilatation of the basilar artery were more obvious. Combined with the above results, the final diagnosis was vertebrobasilar dolichoectasia, right trigeminal neuralgia, and vestibular paroxysmia.

The patient's family refused surgery treatment because of its risk, and drug treatment was chosen. Vitamin B1 (10 mg), methylcobalamin (0.5 mg), and carbamazepine (0.1 g) were given orally 3 times a day to relieve the symptoms. On the third day of drug treatment, the frequency of episodes of paroxysmal vertigo and trigeminal neuralgia was significantly reduced. On the seventh day of drug treatment, the symptoms of paroxysmal vertigo and trigeminal neuralgia were completely relieved, but occasional episodes occurred during the follow-up period. Five

months after discharge, right hemifacial spasm appeared in the patient, which did not affect the quality of life of the patient, so the patient did not choose further treatment. Six months after discharge, the patient was lost to follow-up.

The study was approved by the institutional review board of Harrison International Peace Hospital. Written informed consent was obtained from the patient for publication of this report.

3. Discussion

Vertebrobasilar dolichoectasia is a relatively rare disease of abnormal extension, tortuosity, and dilation of vertebral artery or basilar artery caused by hereditary or autoimmune diseases, accompanied by the hemodynamic changes in the vertebral basilar system. Brain angiography is the gold standard for diagnosing VBD, but MRI can more clearly show the relationship between vascular structure and brain tissue, and is therefore more widely used. In this study, MRI showed that the basilar artery shifted significantly to the right side, reaching the right pontocerebellar peduncle, and the position of basilar artery bifurcation was higher than that of the plane of suprasellar cistern; thus VBD^[2] was confirmed. VBD can clinically induce stroke, brain stem compression, NVCS or hemorrhage, and ahydrocephalus. The most common neurovascular compression syndromes include trigeminal neuralgia (TN; compression of cranial nerve [CN] V), hemifacial spasm (HFS; CN VII), vestibular paroxysmia (VP; CN VIII), and glossopharyngeal neuralgia (GN; CN IX).^[1] The mechanism of those neurovascular compression syndromes is that when the nerve root enters or leaves the brainstem, the nerve roots are compressed by the responsible vessels and show abnormal cranial nerve function. Its clinical manifestations are recurrent episodes of abnormal function, and antiepileptic drugs are usually effective.^[3]

Trigeminal neuralgia is the most common cranial nerve disease, characterized by recurrent episodes of paroxysmal megalgia in the distribution of the trigeminal nerve in 1 side of the face, and the compression of the trigeminal nerve by its

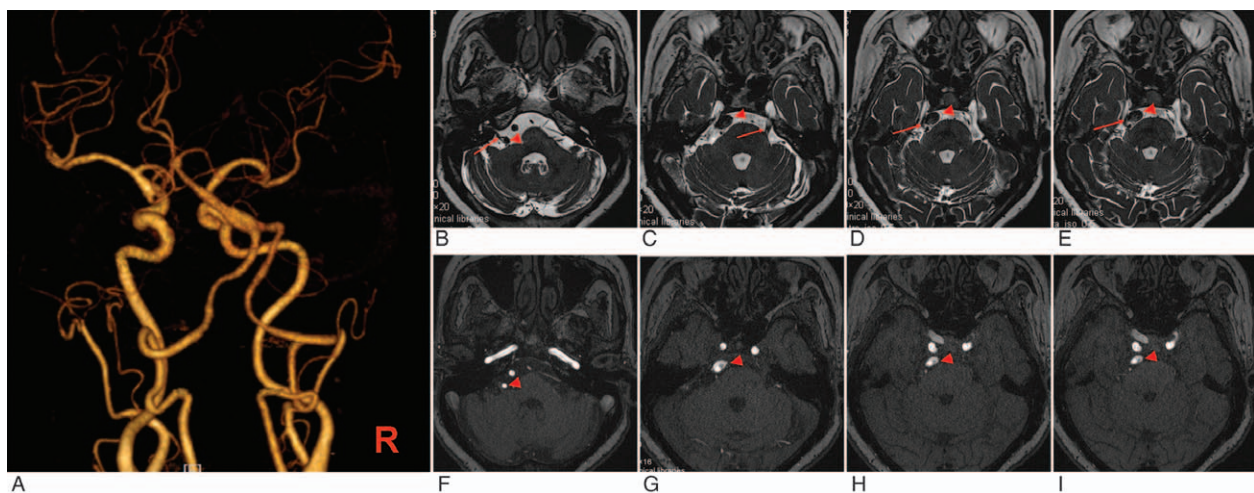


Figure 1. Three-dimensional volume rendering of CTA (A) showed dolichoectasia of the right vertebral artery and basilar artery. Fusion of CISS (B–E, 0.5-mm sections) and TOF angiography sequences (F–I, axial): B and F showed contact between the right vertebral artery (short arrow) and the portion of the cisternal CN VIII (long arrows). The relationship between vestibular paroxysmia and neurovascular compression syndrome was confirmed. No contact between the tortuous vertebral arteries and the cisternal left CN V; D, H, E, and I showed contact between the dilatation of the basilar artery (short arrow) and the superior portion of the cisternal CN V (long arrows). The right CN V was compressed upward and outward by a basilar artery (long arrows). The relationship between vestibular paroxysmia and trigeminal neuralgia was confirmed.

surrounding vessels is the main cause. VP is mainly characterized by transient vertigo, which is caused by vascular compression of the VIII cranial nerve root of the cerebellopontine angle cistern; this is similar to TN. The characteristics of vertigo in this patient included short duration, high frequency, and no significant correlation with body position, which was not consistent with the common causes of vertigo, but was totally consistent with VP.^[4] We did not make a clear diagnosis in the early stage because of the lack of understanding of the disease; meanwhile, the history of trigeminal neuralgia was not given enough attention, and the correlation between trigeminal neuralgia and paroxysmal vertigo had not been paid much attention. Conventional MRI revealed vertebrobasilar artery abnormalities, and the possibility of NVCS was suggested. Three-dimensional reconstructions from 2 types of high-resolution MRI are very useful to create preoperative simulations and determine whether or not to perform operations in TN patients, mainly if associated with VB.^[5] Magnetic resonance 3D-CISS and 3D-TOF-MRA examination showed that the right vertebral artery compressed the right vestibulocochlear nerve, and the dilated basilar artery compressed the right trigeminal nerve. This caused the right trigeminal nerve to move upward and outward; after that, TN and VP were finally confirmed.

The facial nerve and vestibular cochlear nerve have the same cisternal segments and canal segments, both of which originate from the lateral and lower segment of the pons, and pass through the pontocerebellar cistern in oblique angle, then approach the anterior inferior cerebellar artery, and finally enter the inner acoustic meatus. Hemifacial spasm is mainly manifested by involuntary movement of facial muscles, which is also a type of vascular nerve compression syndrome. For our patient, the vestibular nerve was compressed by the right vertebral artery, and the facial nerve may be involved by a close anatomical relationship; therefore, there is a possibility of concurrence with right hemifacial spasm in this patient. It has been confirmed that the right hemifacial spasm occurred 5 months after discharge. In this study, the successive occurrence of trigeminal neuralgia, vestibular paroxysmia, and hemifacial spasm was closely related to VBD; thus progressing VBD was suggested. Male, advanced age, hypertension, smoking, and history of myocardial infarction were independent risk factors for VBD.^[6,7] The physiological changes of VBD are extensive defects in the elastic membrane of the blood vessels and the lack of medial reticular fibers, which resulted in the expansion and tortuosity of the arterial wall under the impact of long-term blood flow. Atherosclerosis induced by hypertension can cause the above physiological changes in the blood vessel wall, thereby accelerating the process.^[6] There is a history of uncontrolled hypertension in this patient, suggesting that it may be related to the progression of VBD.

At present, there is no exact and effective treatment of VBD,^[8] but surgical treatment, such as changes in hemodynamics^[9,10] or direct surgical treatment,^[11,12] is still the most promising treatment. Simple microvascular decompression is effective in the treatment of trigeminal neuralgia caused by VBD.^[13] Antiepileptic drugs are mainly used in medical treatment of NVCS caused by VBD, carbamazepine,^[6] phenytoin, gabapentin, levetiracetam, and other voltage-sensitive sodium channel blockers all effective for those patients.^[14] Although vertigo and neuralgia were well-controlled by carbamazepine,^[15] new symptoms of NVCS continued to occur in our patient. Surgical treatment may be considered when the drug effect is unsatisfactory. At present, microvascular decompression^[16,17] is the most effective surgical treatment. However, the surgical modes,

operation skills, and difficulty of NVCS disease caused by VBD are far different, and the risk is higher than that caused by conventional vascular compression.

4. Conclusion

Comprehensive treatment to control VBD risk factors delays the progression of VBD and improved clinical symptoms may have a better effect on such patients. However, further research is needed.

Author contributions

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