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Case Report

Glossopharyngeal neuralgia with repeated cardiac syncope: A case report

Shunsuke Nakamura, Tatsuya Shimizu, Haruka Tsuneoka, Takaaki Miyagishima, Yuhei Yoshimoto

Department of Neurosurgery, Gunma University, Maebashi, Japan.

E-mail: *Shunsuke Nakamura - gmms0131s@me.com; Tatsuya Shimizu - shimizut@gunma-u.ac.jp; Haruka Tsuneoka - m09201060@gunma-u.ac.jp; Takaaki Miyagishima - miyagisi@gunma-u.ac.jp; Yuhei Yoshimoto - yyoshimo@gunma-u.ac.jp

***Corresponding author:**
Shunsuke Nakamura,
Department of Neurosurgery,
Gunma University, Maebashi,
Japan.

gmms0131s@me.com

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ABSTRACT

Background: Glossopharyngeal neuralgia (GPN) is an uncommon craniofacial pain syndrome. Although rare, it is occasionally associated with cardiac syncope, as vago-glossopharyngeal neuralgia (VGPN).

Case Description: We present the case of a 73-year-old man with VGPN misdiagnosed as trigeminal neuralgia. The patient was diagnosed with sick sinus syndrome, and a pacemaker was introduced. However, syncope still recurred. Magnetic resonance imaging revealed a branch of the right posterior inferior cerebellar artery contacting the root exit zone of the right glossopharyngeal and vagus nerves. We diagnosed VGPN due to neurovascular compression and performed microvascular decompression (MVD). The symptoms disappeared postoperatively.

Conclusion: Diagnosis of VGPN needs appropriate medical interviews and physical examination. MVD is the only curative treatment for VGPN occurring as a neurovascular compression syndrome.

Keywords: Case report, Glossopharyngeal neuralgia, Microvascular decompression, Syncope, Vago-glossopharyngeal neuralgia

INTRODUCTION

Glossopharyngeal neuralgia (GPN) is an uncommon craniofacial pain syndrome characterized by paroxysms of severe stabbing pain unilaterally in the pharynx and ears. Although rare, it is sometimes associated with cardiac syncope and is termed "vago-glossopharyngeal neuralgia" (VGPN). We present a case of GPN with cardiac syncope that was successfully treated by microvascular decompression (MVD).

CASE REPORT

A 73-year-old man presented with an 11-year history of paroxysmal stabbing facial pain. The pain occurred repeatedly and was sometimes associated with loss of consciousness after the pain attack. The patient was referred to a cardiologist for evaluation of the syncope and was diagnosed with sick sinus syndrome. A pacemaker was introduced, although syncope still recurred.

Five years after the first pain episode, the pain progressively worsened. The patient was diagnosed with the right trigeminal neuralgia at a pain clinic and was treated with

carbamazepine and nerve block injection. However, the effect of the treatment was limited, with recurrence of the attacks of pain and loss of consciousness. Although magnetic resonance imaging (MRI) was considered, the pacemaker introduced in the patient precluded performance of MRI. However, after his pacemaker was replaced with an MRI-compatible one during the current year, he was referred to our department for further evaluation and treatment of the facial pain.

The patient's physical/neurological examination revealed no abnormal findings. The pain was located at the right side of the pharynx and the root of the tongue, with radiation to the ipsilateral cheek and ear. It was triggered by swallowing and gargling, with the pain being provoked when food touched his right soft palate while swallowing. The pain was intolerable and lasted for 2–3 min. Chewing or physical stimulation of the face did not cause pain. The patient sometimes lost consciousness after the pain.

MRI revealed a looped branch of the right posterior inferior cerebellar artery (PICA) contacting the root exit zone (REZ) of the right glossopharyngeal and vagus nerves [Figure 1]. There was no arterial contact with the right trigeminal nerve. Organic or functional abnormalities in the cardiovascular system were not observed. Subsequently, the cardiologist diagnosed neuromodulatory syncope. Based on the above, we diagnosed VGPN due to neurovascular compression and performed MVD.

A small 4 cm craniotomy was performed on the right occipital bone with the patient in the park bench position. The looped branch of the PICA was confirmed as compressing the REZ of the glossopharyngeal and vagus nerves from the ventral side [Figure 2]. The PICA was transposed with two Teflon felts. Postoperatively, the pain and syncope disappeared and have currently not recurred as of 9 months after MVD.

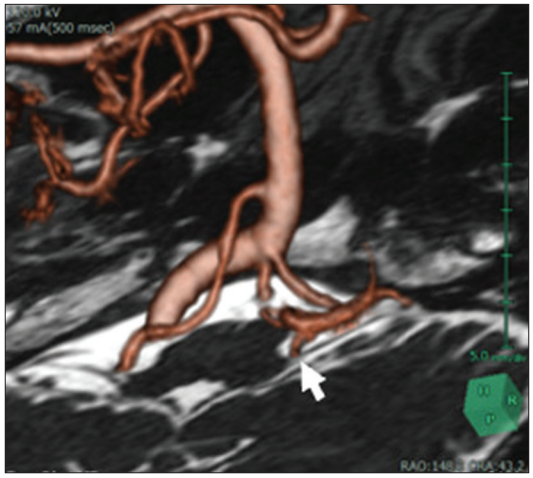

Figure 1: Composite of computed tomography angiography and magnetic resonance imaging constructive interference in the steady state. The right posterior inferior cerebellar artery branch was in contact with the right glossopharyngeal and vagus nerves (arrow).

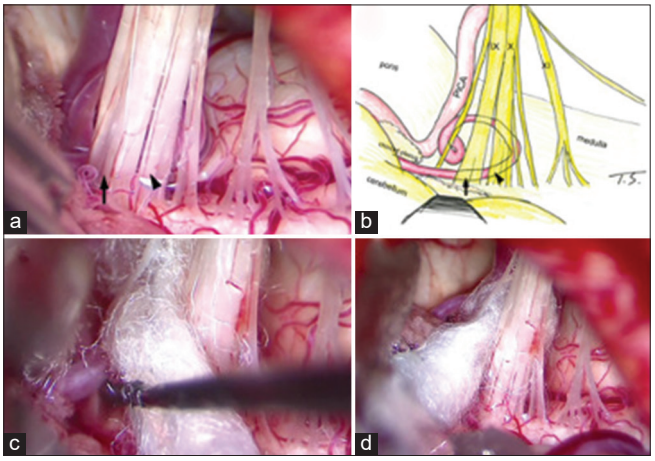

Figure 2: Intraoperative view and schema. (a and b) A branch of the right posterior inferior cerebellar artery (PICA) formed a loop, contacting anteriorly with the root exit zone of the glossopharyngeal (IX, arrow), vagus nerves (X, arrowhead). XI: accessory nerve. (c) The PICA was transposed with Teflon felts and fixed with fibrin glue. (d) Final view.

DISCUSSION

GPN is a rare disease that accounts for 0.2–1.3% of facial pain. The pain begins unilaterally from the root of the tongue and pharynx and radiates to the tympanic membrane, external auditory canal, and auricular region. These locations of pain differ from trigeminal neuralgia.[3,7] Making an accurate diagnosis is quite important because the surgical sites for GPN and trigeminal neuralgia are different. Thus, detailed medical interviews and physical examination are mandatory to differentiate between them.

VGPN accounts for 2% of GPN cases and is a very rare condition.[4,5] The pathogenesis of VGPN is not completely understood, although it is thought to represent a neurovascular compression syndrome of the glossopharyngeal and vagus nerves. Various pathways have been proposed to explain its mechanism: (1) afferent stimulation through the glossopharyngeal nerve to the nucleus of the tractus solitarius reaches the dorsal nucleus of the vagus nerve through a collateral pathway due to the existence of an anatomical relationship between the glossopharyngeal and vagus nerves in the medulla oblongata; (2) afferent stimulation of the glossopharyngeal nerve stimulates the carotid sinus nerve (Hering's nerve); (3) accessory synapses are formed between the glossopharyngeal and vagus nerves in the regions of their ganglia; and (4) there is an abnormal communication involving the solitary nucleus and the nucleus ambiguus.

In any case, the stimulated vagus nerve activates the vagus nerve-carotid baroreceptor pathway, resulting in marked bradycardia (cardioinhibitory effect) and reduction in peripheral vascular resistance (vasodilatory effect), which ultimately provoke syncope.[2] In VGPN, not only bradycardia and cardiac arrest associated with parasympathetic activation but also hypotension with vasodilation due to inhibition of sympathetic activity are thought to be involved.[6] Temporary or permanent pacemaker therapy does not prevent syncopal episodes when peripheral vascular resistance is severely attenuated.[4] Therefore, it is thought to be difficult to treat syncope in VGPN using pacemaker therapy alone.

In addition, carbamazepine, which is used in the medical treatment of neuralgia, produces side effects such as bradyarrhythmia and atrioventricular block.[1] Thus, it can also cause drug-induced syncope. MVD is currently considered the only curative treatment for both GPN and syncopal episodes and has, so far, been indicated for GPN cases who have not responded to medical therapy. However, in cases of VGPN, MVD should be considered at an early stage once a reliable diagnosis is made.

CONCLUSION

MVD is considered to be the only curative treatment for VGPN. Although diagnosis of VGPN is not always easy, early diagnosis by appropriate medical interviews and physical examination is important.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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