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Case Report

A Case of Vago-Glossopharyngeal Neuralgia Caused by Choroid Plexus

Abstract

Vascular compression has been reported to be the most common reason for vago-glossopharyngeal neuralgia (VGN). The treatment may include medications, ganglion blockade with a radiofrequency ablation, and microvascular decompression (MVD). A review of the literature reveals that VGN may develop due to choroid plexus compression, and the number of reported cases is very limited. The current case is the fifth in the relevant literature. In this paper, choroid plexus compression has been shown intraoperatively during the treatment of rare idiopathic VGN using MVD. Complaints of the patient have been resolved following the choroid plexus excision.

Keywords: *Choroid plexus, microvascular decompression, vago-glossopharyngeal neuralgia*

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Introduction

Vago-glossopharyngeal neuralgia (VGN) is a rare craniofacial syndrome. Typically, paroxysmal electricity, shock-like neuropathic pain is felt in innervation fields of glossopharyngeal nerve and pharyngeal branch of vagus. These fields usually extend from the ear to gonion and therefore, root of the tongue, soft palate, lateral, and posterior pharyngeal wall are affected by the pain,[1,2] which is identified as unilateral. Talking, coughing, or yawning commonly trigger this pain and burn sensation is described after the pain. In addition, patients usually lose weight as pain is also triggered by chewing and swallowing. Its yearly incidence is 0.7/100,000; it is more common in patients in their 50s and above.[3] VGN could have idiopathic or secondary reasons, such as vascular compression, which is the most commonly, reported idiopathic reason for VGN, and choroid plexus compression, which has been reported as one of the secondary reasons.[4] However, choroid plexus compression-related VGN is a very rare condition reported in the literature to date. In this paper, choroid plexus compression on root entry zones of 9th and 10th nerves detected intraoperatively during treatment of VGN with microvascular decompression (MVD) is reported.

Case Report

A 64-year-old female patient had been diagnosed with left VGN 4 years before her admission to our clinic and had been treated with carbamazepine, and gabapentin, which are effective on neuropathic pain. Ganglion blockade with radiofrequency ablation (RFA) had been used twice; as a result, the patient had benefited from these procedures for a short time. However, on her admission, she had an electricity-shock-like intolerable pain beginning from the left ear and extending to the larynx. She stated that talking and chewing triggered the pain. The case of the current study described the pain she had as almost the same as the other patients of VGN reported to have in the relevant literature. In other words, no other characteristics of pain caused by choroid plexus compression were described by our patient compared to typical pain described by VGN patients. Moreover, she had neuralgia-related sleepiness, weight loss, and depression. On her cranial magnetic resonance imaging (MRI) [Figure 1], there was an image suspected as a vascular decompression. The patient underwent an MVD operation and her pain relieved in postoperative period. When the images of MRI [Figure 2] were analyzed retrospectively, we noticed that choroid plexus could be observed on the images of MRI although it was shown in grey.

MVD was applied to the 9th and 10th cranial nerves of the patient with the left retrosigmoid lateral suboccipital

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craniotomy on prone position under microscope. A significant posterior inferior cerebellar artery (PICA) compression was not observed from anterior on the 9th and 10th cranial nerves in intraoperative observation; on the other hand, choroid plexus tissue causing compression on root entry zones [Figure 3a] was identified and it was resected. Afterward, root entry zone compression was seen to disappear, and a clear change of the color of the root entry zones of the 9th and the 10th cranial nerves was observed [Figure 3b]. Despite the absence of a significant vascular compression, PICA, and the 9th and the 10th cranial nerves were separated with a Teflon sponge. In the postoperative period, the patient had no pain, which was reported to exist before the operation, related to VGN. No complications were detected in the postoperative course. She had no pain or any other complaints at the end of the 6th month after the surgery.

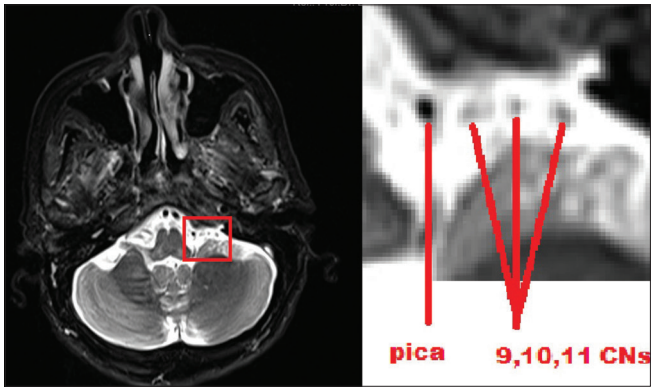

Figure 1: Preoperative T2 axial magnetic resonance imaging image shows posterior inferior cerebellar artery compression on the 9th and 10th cranial nerves

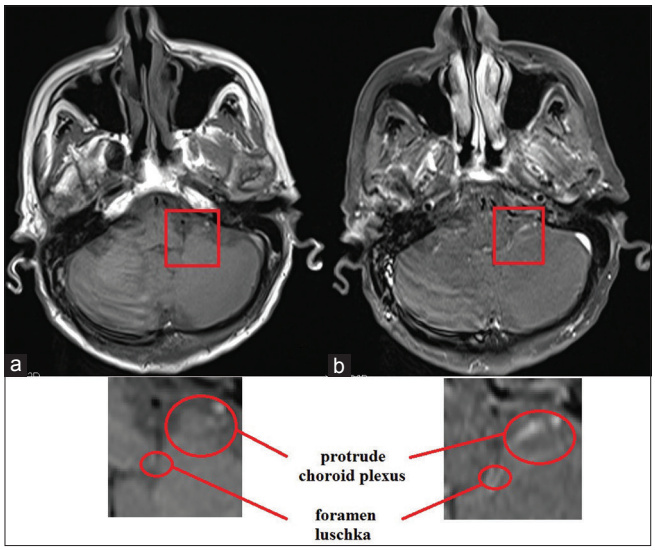

Figure 2: Preoperative T1 axial magnetic resonance images, both non-contrast enhanced (a) and contrast enhanced (b) images, showing choroid plexus tissue protruding from foramen of Luschka

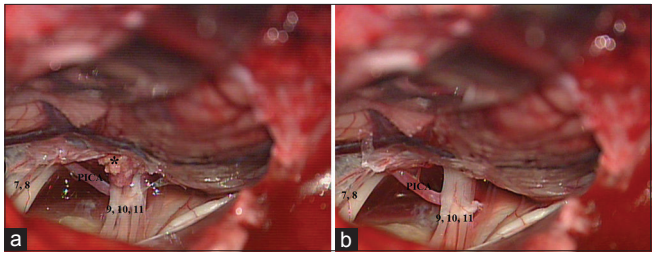

Figure 3: (a) The position of posterior inferior cerebellar artery and choroid plexus compression on root entry zone of the 9th and 10th nerves are demonstrated in intraoperative microscope image. *: Choroid plexus. (b) The color change of the root entry zones of the 9th and the 10th cranial nerves is demonstrated following the excision of the choroid plexus

Discussion

Compared to trigeminal neuralgia, which is the most common cranial rhizopathy, the incidence rate of VGN is quite less. This ratio varies between 5.6:1 and 100:1.[5,6]

Vascular compression is the most common reason for VGN. Secondary reasons could include neoplasms, vascular malformations, demyelinating diseases (multiple sclerosis), infection, trauma, Chiari malformation, Eagle's syndrome, and choroid plexus overgrowth.[4] A case of choroid plexus compression-related VGN was first described by Occhiogrosso *et al*. in 1980[7] and they reported a series of four cases in 1996. In their series, lateral choroid plexus located in the 4th ventricle was described to protrude from foramen of Luschka and caused VGN by making a compression on the 9th and 10th nerves.[8] Two VGN cases related to choroid plexus papilloma compression together with vascular compression were reported by Greene *et al*.[9] A very rare case of VGN related to a lipoma located in cerebellopontine angle was reported by Choi *et al*.[10] This type of atypical conditions should also be considered while the images of MRI are analyzed and evaluated for VGN.

Treatment options for VGN include rhizotomy with RFA, gamma knife, and MVD.

Conclusion

In the current case, the patient, whose MRI images preoperatively suggested a vascular compression, was operated with prediagnosis of idiopathic VGN and a compression of choroid plexus tissue on the root entry zone was observed intraoperatively, which is a very rare condition reported in the relevant literature. The condition was resulted from extrusion of choroid plexus tissue in the 4th ventricle from foramen of Luscka.[8] To the best of our knowledge, this is the fifth VGN case related to choroid plexus compression in the literature.

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Conflicts of interest

There are no conflicts of interest.

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