Microvascular Decompression Alone without Rhizotomy Is an Effective Way of Treating Glossopharyngeal Neuralgia: Clinical Analysis of 46 Cases

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Keywords

Glossopharyngeal neuralgia · Microvascular decompression ·
Glossopharyngeal nerve rhizotomy · Vagal nerve rhizotomy ·
Functional neurosurgery

Abstract

Background: Microvascular decompression (MVD) has been the right choice for glossopharyngeal neuralgia (GPN) patients. However, whether glossopharyngeal/vagal nerve root rhizotomy should be combined with MVD is still controversial. **Objective:** To evaluate whether glossopharyngeal/vagal nerve root rhizotomy during MVD is necessary for the treatment of GPN. **Methods:** We performed a retrospective study of 46 GPN patients who underwent MVD surgery alone in our hospital, and their patient demographics, clinical presentations, and intraoperative findings are shown. The immediate and long-term follow-up outcomes were investigated to show the treatment's efficiency and safety; the outcome was also compared with our previous study. The relevant literature was reviewed to show complications for

GPN patients undergoing glossopharyngeal/vagal nerve root rhizotomy with MVD. **Results:** The most common of-fending vessel was the posterior inferior cerebellar artery (60.9%). 100% of the patients were pain-free (score of I on the Barrow Neurological Institute pain intensity [BNI-P] scale) immediately after MVD surgery, while 1 patient re-lapsed with occasional pain 12 months after the operation (score of III on the BNI-P scale). Poor wound healing and hearing loss were found in 1 case each. No complications re-lated to the glossopharyngeal nerve/vagal nerve were re-ported. Some surgical techniques, such as thorough explo-ration of the CN IX–X rootlets, full freeing from arachnoid adhesions, and usage of a moist gelatin sponge, can improve the success rate of the operation. **Conclusions:** MVD alone without rhizotomy is an effective and safe method for pa-tients with GPN.

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Introduction

Glossopharyngeal neuralgia (GPN) [1, 2] is a rare cranial nerve hyperactivity pain syndrome, which was first described by Weisenburg [3] in 1910 in a patient with a cerebellopontine angle tumor. Patients with GPN usually experience repeated, sharp, severe, electric-shock-like facial pain in the base of the tongue, ear, tonsillar fossa, and/or submandibular region [4–6], lasting from a few seconds to minutes. The pain can be precipitated by swallowing, talking, coughing, or chewing, thus seriously affecting patients' quality of life.

The first-line treatment for GPN is pharmacological therapy, including mainly anticonvulsant medications such as carbamazepine, oxcarbazepine, gabapentin, and pregabalin [6, 7]. However, if patients are refractory/resistant to medicine, or having drug allergies or unbearable side effects, surgical treatment may be considered.

Several potentially successful maneuvers are available for patients with medically intractable GPN, such as microvascular decompression (MVD) with or without rhizotomy, Gamma Knife radiosurgery, stereotactic radio-

surgery, and percutaneous glossopharyngeal nerve block. Among these, MVD has turned out to be a good choice for idiopathic GPN since Jannetta introduced MVD for GPN in 1981 [8]. However, whether glossopharyngeal/vagal nerve root rhizotomy should be combined with MVD is still controversial.

In this study, we present a series of 46 patients who underwent MVD alone without rhizotomy during 2013–2018 in our hospital. The immediate and long-term follow-up outcomes were investigated to show the treatment's efficiency and safety. We also compared the results with our previous research [9] and found that the patients in this study had a better prognosis. We analyzed the differences and propose several vital points during surgery to ensure its efficacy.

Subjects and Methods

Patient Selection and Pre- and Postoperative Evaluation

Between April 1, 2013, and December 31, 2018, 49 consecutive patients suffering from medically intractable GPN underwent an operation in our hospital. The records on these patients

Fig. 1. MRI of a typical right-sided glossopharyngeal neuralgia patient showing that the right posterior inferior cerebellar artery was adjacent to the root entry zone of the glossopharyngeal nerve, which caused the symptoms. **a** Three-dimensional coherent oscillatory state acquisition for the manipulation imaging contrast (3D-

COSMIC). **b** Three-dimensional time-of-flight magnetic resonance angiography (3D-TOF-MRA). The solid red arrows indicate the posterior inferior cerebellar artery; the empty red arrows indicate the vertebral artery.

were accessed and evaluated by two researchers. After excluding 2 patients with secondary GPN related to meningiomas and 1 patient with recurrent GPN who underwent his first MVD surgery in other hospital, all remaining 46 patients met the diagnostic criteria for GPN according to the International Classification of Headache Disorders, 3rd edition [6] (Table 1), underwent MVD without rhizotomy, and were included in this retrospective study.

Preoperative 3.0-T MRI examinations were performed to detect neurovascular compression and to rule out other diseases (Fig. 1). The Barrow Neurological Institute pain intensity (BNI-P) score [10] (I: no pain, no medication; II: occasional pain, not requiring medication; III: some pain, adequately controlled with medication; IV: some pain, not adequately controlled with medication; and V: severe pain/no pain relief) and facial numbness (BNI-N) score [10] (I: no facial numbness; II: mild facial numbness, not bothersome; III: facial numbness, somewhat bothersome; and IV: facial numbness, very bothersome) were assessed to evaluate the severity of the neuralgic pain and the postoperative outcome.

Surgical Procedure

Under general endotracheal anesthesia, all patients underwent MVD via a conventional retrosigmoid approach in a park bench position. Brainstem auditory evoked potentials were assessed intraoperatively to avoid any hearing disturbance. The lower cranial nerves were fully revealed under an operative microscope after dissection of the arachnoid membrane of the cerebellopontine angle

and gentle retraction of the cerebellum. The full length of the glossopharyngeal and vagal nerve, including the root entry zone, was explored appropriately and progressively to identify all offending blood vessels. Small pieces of a moist gelatin sponge, Teflon felt, and a moist gelatin sponge again were placed between the vessels and nerves as a sandwich-like complex (Fig. 2). Surgeons should minimize any traction on the glossopharyngeal and vagal nerves and reconfirm that the nerves are free from any offending vessels, whether they are arteries or veins.

Table 1. Diagnostic criteria for glossopharyngeal neuralgia

A. Recurring paroxysmal attacks of unilateral pain in the distribution of the glossopharyngeal nerve ¹ and fulfilling criterion B
B. Pain has all of the following characteristics:
1. lasting from a few seconds to 2 min
2. severe intensity
3. electric shock-like, shooting, stabbing, or sharp in quality
4. precipitated by swallowing, coughing, talking, or yawning
C. Not better accounted for by another ICHD-3 diagnosis

¹ Within the posterior part of the tongue, tonsillar fossa, pharynx, or angle of the lower jaw and/or in the ear.

Fig. 2. Intraoperative photos showing the “moist gelatin sponge/Teflon felt/moist gelatin sponge” complex. * Moist gelatin sponge; Δ Teflon felt. PICA, posterior inferior cerebellar artery.

For patients with GPN and trigeminal neuralgia (TN), we dealt with the lower cranial nerve-vascular complex first, then explored the trigeminal nerve and decompressed the offending vessels related to it.

Follow-Up and Statistical Analysis

Follow-up data were obtained by telephone interview or outpatient visit. Surgical outcomes, including the degree of pain (BNI-P score), pain recurrence, and postoperative complications, were collected.

Continuous variables are reported as the mean $\pm$ standard deviation, and categorical variables are expressed as n (%). All statistical analyses were performed using R statistical software version 3.5.1 (The R Foundation for Statistical Computing, Vienna, Austria). A p value <0.05 was regarded as statistically significant.

Results

Patient Demographics and Clinical Presentation

Of the 46 patients with GPN in this study, 20 (43.5%) were male; the mean age at the time of surgery was 61.6 ± 11.8 years. On average, the patients had been symptomatic for 43.1 ± 62.3 months before surgery (range, 0.25–252), and more patients presented with left-sided symptoms (56.5%; 26 of the 46 patients) than with right-sided symptoms. The most common symptom was pharynx pain, followed by tongue pain. There were more patients with symptoms at a single site (27; 58.9%) than with symptoms at multiple sites (19; 41.3%). Six patients (13.0%) experienced TN complicated by GPN. One patient experienced cardiac arrhythmias and syncope during pain episodes, and another patient experienced syncope only. Of the 46 patients with GPN, 15 (32.6%) scored IV and 31 (67.4%) scored V on the BNI-P scale and no one had facial numbness before the operation (Table 2).

Intraoperative Findings

In this study, neurovascular compression was found in all 46 patients during the operation. The most common offending vessel was the posterior inferior cerebellar artery (PICA), which was responsible for the neurovascular compression in 28 (60.9%) of the 46 patients, followed by the vertebral artery (VA) + PICA (15.2%), PICA + anterior inferior cerebellar artery (6.5%), anterior inferior cerebellar artery (6.5%), VA (4.3%), small arteries (4.3%), and PICA + vein (2.2%) (Table 3).

Six patients (13.0%) experienced TN complicated by GPN. All of them underwent glossopharyngeal, vagal, and trigeminal nerve decompression, and the superior cerebellar artery was responsible for their TN symptoms.

Table 2. Patient demographics and clinical presentation ($n = 46$)

Gender	
Male	20 (43.5%)
Female	26 (56.5%)
Mean age $\pm$ SD, years	61.6 $\pm$ 11.8
Mean duration of symptoms to surgery $\pm$ SD, months	43.1 $\pm$ 62.3
Side of lesion	
Left	26 (56.5%)
Right	20 (43.5%)
Distribution of symptoms	
Pharynx only	18 (39.1%)
Tongue only	5 (10.9%)
Ear only	4 (8.7%)
Pharynx + tongue	5 (10.9%)
Pharynx + ear	5 (10.9%)
Tongue + tonsillar fossa	1 (2.2%)
Tongue + lower jaw	1 (2.2%)
Other complex symptoms	7 (15.2%)
Uncommon manifestations	
Neurocardiogenic syncope	2 (4.3%)
Cardiac arrhythmias	1 (2.2%)
Asystole	0 (0%)
Convulsive crises	0 (0%)
Glossopharyngeal neuralgia with trigeminal neuralgia	6 (13.0%)
BNI-P score	
IV	15 (32.6%)
V	31 (67.4%)
BNI-N score	
I	46 (100%)
Mean follow-up duration $\pm$ SD, months	32 $\pm$ 17.9

The heart rate of 1 patient dropped to a minimum of 30 beats/min during vessel decompression; atropine was used, and the operation was suspended until the heart rate rose again. The patient who suffered from cardiac arrhythmias (sometimes sinus arrest) and syncope during pain episodes had a cardiac pacemaker implanted before MVD surgery, and no particular events occurred during the operation.

Immediate and Follow-Up Postoperative Outcomes

There was no mortality due to or related to the operation in this group. All the 46 patients could be contacted for follow-up data, with a mean follow-up duration of 38 ± 17.9 months (range, 12–80). 100% of the patients were pain-free (score of I on the BNI-P scale) immediately after the MVD surgery, while one 62-year-old female patient relapsed with occasional pain 12 months after the operation. She retook carbamazepine, and the pain could be controlled well (score of III on the BNI-P scale).

Table 3. Distribution of offending vessels

Offending vessels	Patients
PICA with/without its branches	28 (60.9%)
VA + PICA	7 (15.2%)
PICA + AICA	3 (6.5%)
AICA with/without its branches	3 (6.5%)
VA	2 (4.3%)
Small arteries	2 (4.3%)
PICA + vein	1 (2.2%)

PICA, posterior inferior cerebellar artery; VA, vertebral artery; AICA, anterior inferior cerebellar artery.

Table 4. Comparison between our previous and the present study

	Previous study [9]	Present study	<i>p</i> value
Gender (male:female), <i>n</i>	15:20	20:26	0.95
Mean age at surgery ± SD, years	56.8±8.1	61.6±11.8	0.04*
Side (left:right), <i>n</i>	19:16	26:20	0.84
Relief rate			
Immediate	94.3% (33/35)	100% (46/46)	0.18
1-month follow-up	88.6% (31/35)	100% (46/46)	0.03*
1-year follow-up	82.6% (29/35)	97.8% (45/46)	0.04*

Analysis of variance was used for continuous variables, Pearson's χ^2 test or Fisher's exact test for categorical variables. * *p* < 0.05.

In this study, 6 patients (13.0%) experienced TN complicated by GPN. All of them underwent glossopharyngeal, vagal, and trigeminal nerve decompression, and the superior cerebellar artery was responsible for their TN symptoms. One patient complained of facial numbness (score of III) immediately after the operation, but the symptom gradually relieved in 6 months.

Poor wound healing was found in 1 case, which subsequently healed after debridement. Hearing loss occurred in 1 case, not fully recovering within the 6-month follow-up period; the recovery situation is still being observed. No dysphagia, dysphonia, cerebrospinal fluid leak, facial palsy, hoarseness, dystaxia, or drinking cough occurred during follow-up.

Comparison with Our Previous Study

In 2017, we had reported a case series of 35 GPN patients who underwent MVD surgery alone in our hospital between 2004 and 2006 by Dr. Zhao [9]. In this early

period, we used Teflon felts alone instead of Teflon with a moist gelatin sponge as a sandwich-like complex in the decompression process. Furthermore, we had not emphasized any full-length dissection of the CN IX–X rootlets or thorough freeing from arachnoid adhesions. The outcomes, compared with those of our present study, are shown in Table 4. Although there was no significant difference in the rate of immediate pain relief between the two studies, the 1-month and 1-year follow-ups revealed that our present operation method is the better choice.

Discussion

GPN, also known as vagoglossopharyngeal neuralgia, is a rare clinical neurological disorder characterized by paroxysmal craniofacial pain. This disease was first described by Weisenburg in 1910 in a male patient with a right pontocerebellar angle tumor [3] and was named “essential velopharyngeal pain” at that time. The current name, “glossopharyngeal neuralgia,” was introduced by Harris in 1921 [11]. As the glossopharyngeal nerve is near to the vagal nerve, some patients simultaneously show symptoms of the vagal nerve, such as bradycardia, syncope, or even cardiac arrest, as well as the craniofacial pain. Thus, some scholars believe that this condition may be more appropriately named “vagoglossopharyngeal neuralgia” [12]. In this article, we still use the term “glossopharyngeal neuralgia,” since we focused mostly on pain.

The first-line treatment for GPN is pharmacologic [6, 7, 13], but neurosurgical approaches should still be considered in cases of severe or medication-refractory GPN. Of all surgical treatments, MVD may be proposed as the primary neurosurgical option due to its high relief rate – which has often been higher than 90% in more recent series – and excellent long-term outcome [12, 14, 15]. In comparison, other treatments such as Gamma Knife radiosurgery lead to early recurrence [16, 17], and percutaneous radiofrequency thermocoagulation has a high incidence of side effects and therefore has not gained full acceptance; thus, both are not as versatile as MVD.

Some researchers advocated MVD with glossopharyngeal/vagal nerve rhizotomy as an excellent method and insisted that destruction of the glossopharyngeal nerve/vagal nerve does not lead to serious neurological deficits [18]; however, further studies showed complications such as dysphagia, hoarseness, and swallowing disturbance might occur at a certain rate [19, 20].

Wang et al. [19] reported on a series of 37 patients with GPN; 22 of them underwent MVD and 15 underwent MVD plus glossopharyngeal nerve root rhizotomy. Between the two groups there was no significant difference in postoperative efficacy, while the incidence of postoperative hoarseness and drinking cough in the MVD + GNR group was higher than in the MVD group. Kim et al. [21] also had the same idea: they performed MVD surgery alone on 30 patients with intractable primary typical GPN, and 29 of the 30 patients (96.7%) achieved excellent results and no persistent morbidity was reported.

In this study, 46 patients with medically intractable primary GPN who underwent MVD alone without rhizotomy were studied. All of them attained immediate pain relief after surgery, and 97.8% (45 of 46) of them achieved an excellent prognosis during a 1-year follow-up. We also previously reported on a series of 30 patients with long-term follow-up (mean 132 months) [9], whose outcome was also favorable. No complications related to the glossopharyngeal nerve/vagal nerve were reported in these two studies. Therefore, we hold the opinion that MVD alone is an effective and safe method for GPN patients. Now that the simple decompression process is effective, it is not necessary to carry out rhizotomy.

Moreover, we compared patients' outcomes between our two studies. Due to the aging of the population, the patients in this study were older than the previous ones, while the other baseline data did not display any statistically significant difference. Although the results of the two studies are both satisfactory, we found significant improvement in the 1-month and 1-year follow-up in the current trial (Table 4). Our previous article in 2017 reported on patients with GPN who underwent MVD surgery between January 2004 and June 2006, while the present study reports on 46 new patients treated between April 2013 and December 2018. The patients enrolled in this study were operated on nearly 10 years later than those in our last study. Over the course of this decade, we had learned lessons from over 10,000 MVD operations for hemifacial spasm (HFS) and then integrated and summed up a set of practical methods. Therefore, there is every reason to believe that the pain remission rate with MVD depends on the surgeon's experience and other technical tips to some extent.

First of all, one of the critical points of MVD surgery, be it for HFS, TN, or GPN, is full-length decompression. Both the root entry zone and the distal part of the nerves should be adequately decompressed; all vessels, including arteries, veins, and their perforating branches, must be removed from the nerves. For GPN patients, we explored

the VA, PICA, perforating branches, and veins sequentially.

Some scholars [19] believe that the pathogenesis of GPN is due to neurovascular compression, accompanied by local thickening and adhesion of the arachnoid membrane, which leads to demyelination of the glossopharyngeal nerve and vagal nerve roots, resulting in a short circuit between the nerve fibers which triggers the onset of pain. Other studies also reported several patients with GPN caused by an adhesive arachnoid [22, 23]. Thus, from our point of view, the thickening and adhesion of the arachnoid should be separated and loosened entirely.

The recurrence rate following MVD was reported to be about 7% [24]. Palanisamy et al. [25] reported the case of a recurrent GPN patient who underwent MVD three times. Two small arteries, one above the glossopharyngeal nerve, another below the vagal nerve, and arachnoid adhesions found in the lateral cerebellomedullary cistern were responsible for the third exploration surgery. Therefore, we emphasize again the importance of a thorough exploration of the glossopharyngeal/vagal nerve and full separation of the arachnoid membrane to avoid failure or recurrence with MVD surgeries in idiopathic GPN. In this study, we also used a "moist gelatin sponge/Teflon felt/moist gelatin sponge" complex instead of Teflon felt alone to decompress the nerves. Using this sandwich-like complex can reduce postoperative adhesions according to our previous experience on HFS and TN patients.

Although MVD seems to be a first-line surgical method, there still have been some complications reported, such as short-term hoarseness and drinking cough, cerebrospinal fluid leakage, intracranial infection, and ipsilateral hearing loss [19]. Limitations of this study include a relatively small sample size, short follow-up period, and retrospective design. We will further analyze these issues as the number of patients admitted and the follow-up time increase.

Conclusion

MVD alone without rhizotomy is an effective and safe method for patients with GPN.

Statement of Ethics

All of the procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional research committee. Informed and written consent was obtained from all the individual participants included in this study.

Disclosure Statement

The authors have no conflict of interest to declare.

Funding Sources

No funding was received for this study.

Author Contributions

X. Zheng, X.-Y. Wei, and S.-T. Li conceived and designed this study; Y. Yuan and T.T. Ying performed the electrophysiological monitoring; X. Zheng, J. Zhu, and X.-Y. Wei provided the material and collected the clinical data; X. Zheng and J. Zhu analyzed the data. All authors gave their final approval for the submission of this paper for publication.

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