ISSN 1738-6217
J Kor Soc Ster Func Neurosurg 2017;13(1,2):15-19

CASE REPORT

The Treatment of Combined Trigeminal and Glossopharyngeal Neuralgia by Rhizotomy : A Case Report

Sung Hyun Noh, MD[1], Sang Sup Chung, MD, PhD[2], Joo Pyung Kim, MD, PhD[2]

[1]1Department of Neurosurgery, Gangnam Severance Hospital, Yonsei University College of Medicine, Seoul, Korea
[2]Department of Neurosurgery, Bundang Medical Center, CHA University, Seongnam, Korea

Combined trigeminal and glossopharyngeal neuralgia, which is most often caused by vascular compression at the root entry zone of the cranial nerves, is an exceedingly rare cranial neuralgia. In our case, a 57-year-old woman underwent a rhizotomy for combined trigeminal and glossopharyngeal neuralgia in the absence of vascular compression. No recurrence was observed in follow-up examinations. We report an excellent clinical outcome and discuss the clinical presentation and surgical treatment.

KEY WORDS: Rhizotomy · Cranial neuralgia · Trigeminal and glossopharyngeal neuralgia.

INTRODUCTION

Combined trigeminal and glossopharyngeal neuralgia is a rare cranial neuralgia. In 1930, Filatov, et al.[1)] reported the first case of combined trigeminal and glossopharyngeal neuralgia. This condition accounts for 10−46.7% of glossopharyngeal neuralgia cases and 0.3−0.5% of trigeminal neuralgia cases.[1)] In 1981, Rushton, et al.[1)] reported 217 cases of glossopharyngeal neuralgia at the Mayo Clinic between 1922 and 1977. Out of the 217 cases, there were 25 cases of combined trigeminal and glossopharyngeal neuralgia14. In most cases, an artery and vein compressed the root entry zones of the trigeminal and glossopharyngeal nerves in trigeminal and glossopharyngeal neuralgia.[2)] Therefore, microvascular decompression (MVD) is a widely accepted surgical therapy with a high success rate (80%), good long-term outcome, and the fewest side effects. We report the case of a 57-year-old woman who underwent a rhizotomy for combined trigeminal and glossopharyngeal neuralgia in the absence of vascular compression. The clinical presentation, surgical treatment, clinical outcome, and relevant literature are discussed.

Address for correspondence: Joo Pyung Kim, MD, PhD
Department of Neurosurgery, Bundang CHA Medical Center, CHA University College of Medicine, 59 Yatap-ro, Bundang-gu, Seongnam 13496, Korea
Tel: +82-31-780-5688, Fax: +82-31-780-5269
E-mail: tsmek@hanmail.net

CASE REPORT

History and presentation

A 57-year-old female with a 12-year history of lancinating pain in the right cheek and auricle consistent with trigeminal neuralgia presented to our hospital. Symptoms was sometimes sudden, severe, stabbing, lightning-like, and shock-like pain attacks that are usually one-sided in the 2nd and 3rd trigeminal branch. She had severe, transient, sharp pain in the base of the right tongue and the tonsillar fossa when swallowing, consistent with glossopharyngeal neuralgia. The pain was so severe that the patient found it hard to endure, making it difficult to swallow. Her initial symptom was glossopharyngeal pain and then after 5 years, trigeminal pain developed. She had been treated with carbamazepine, oxcarbazepine, and gabapentin for 12 years, but the symptoms were poorly controlled. Her pain was rated as V scale on The Barrow Neurological Institute (BNI) Pain Intensity Scale.

Examination

We used magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) to find the offending vessel. A 1.5 Tesla machine (Philips MR System Achieva, Netherlands) acquiring continuous slices of 1-mm thickness was used for MRI. No artery or vein appeared to cause the trigeminal neuralgia, and no artery or vein traversed the glossopharyngeal nerve (Fig. 1). For finding trigger zones, we sprayed lidocaine onto the tonsillar fossa and the pain decreased. Based on clinical symptoms and examinations, we

confirmed a diagnosis of combined trigeminal and glossopharyngeal neuralgia.

Operation and outcome

A lateral retrosigmoid suboccipital approach was performed, and a careful dissection of the arachnoid membrane along the entire length of the trigeminal nerve root was performed from the ventral aspect of the pons to the entrance of Meckel's cave. During exploration, we found no vascular compression of the trigeminal nerve in the operative field. We performed a rhizotomy of a quarter of the sensory part of the trigeminal nerve in its caudolateral aspect approximately 3mm from the pons (Fig. 2). Then, we dissected the arachnoid membrane of the entire 9−11th nerve complex. After we were unable to find an offending vessel, we performed a rhizotomy of the glossopharyngeal nerve and the upper rootlet of the vagus nerve (Fig. 3). During the operation, we used a micro-dissector to touch the 9−11th nerve complex and found the parasympathetic root. Based on the resulting sinus bradycardia, we found the upper rootlet of the vagus nerve and then performed a rhizotomy of the glossopharyngeal nerve and the upper rootlet of the vagus nerve. There were no complications such as hoarseness or dysphasia after the operation. Three months after the surgery, the patient appealed no symptoms. The BNI Pain Intensity Scale score of the patient's pain was reduced to II. There was no symptom at follow-up three years later.

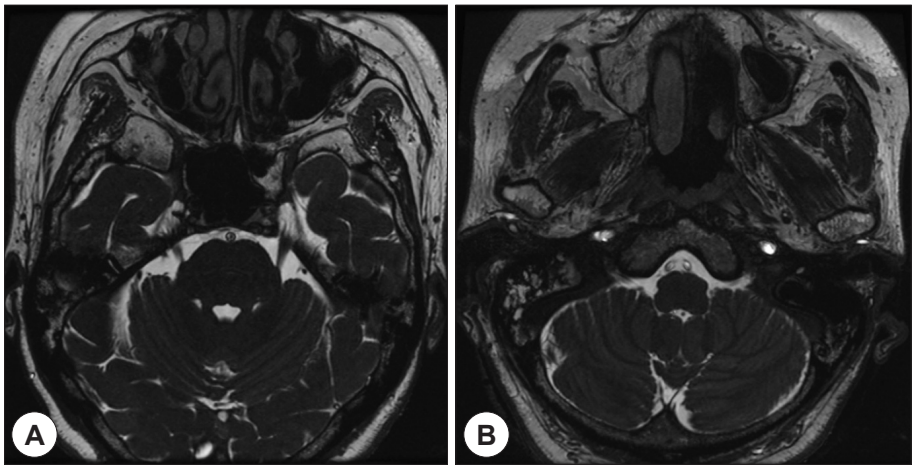

Fig. 1. No artery or vein appeared to cause the trigeminal neuralgia, and no artery or vein traversed the glossopharyngeal nerve in magnetic resonance imaging.

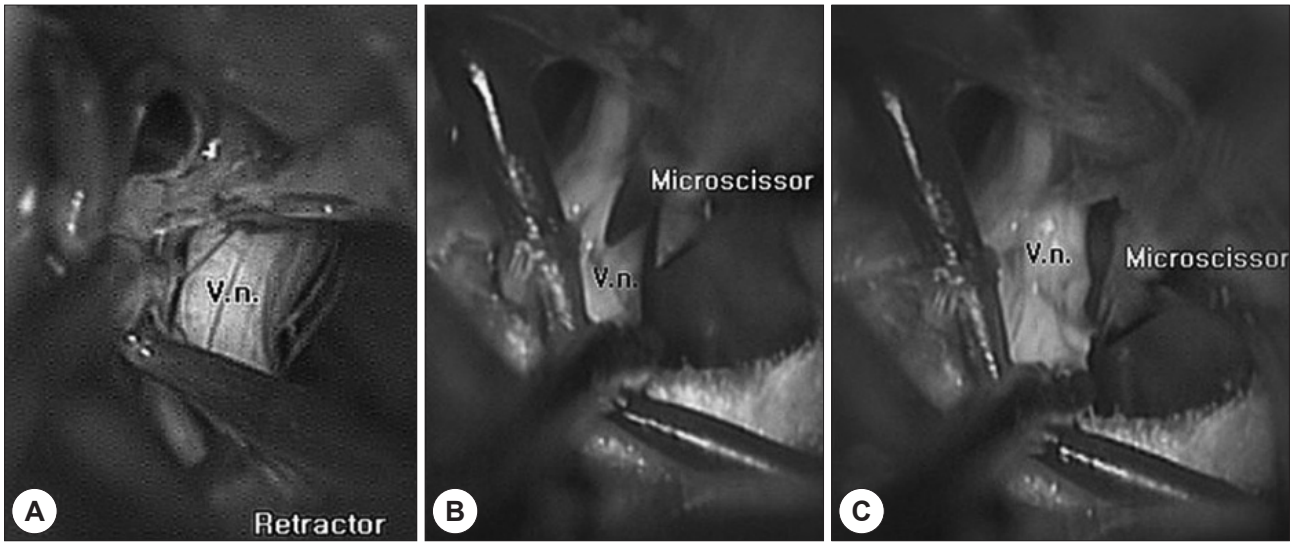

Fig. 2. We dissected the arachnoid membrane along the entire length of the trigeminal nerve to attempt to isolate the vascular compression in the proximal or distal parts of the trigeminal nerve (A). We retracted the vein with a micro-dissector for the rhizotomy of the sensory part of the trigeminal nerve root (B). We performed a rhizotomy of a quarter of the sensory part of the trigeminal nerve in its caudolateral aspect approximately 3 mm from the pons (C).

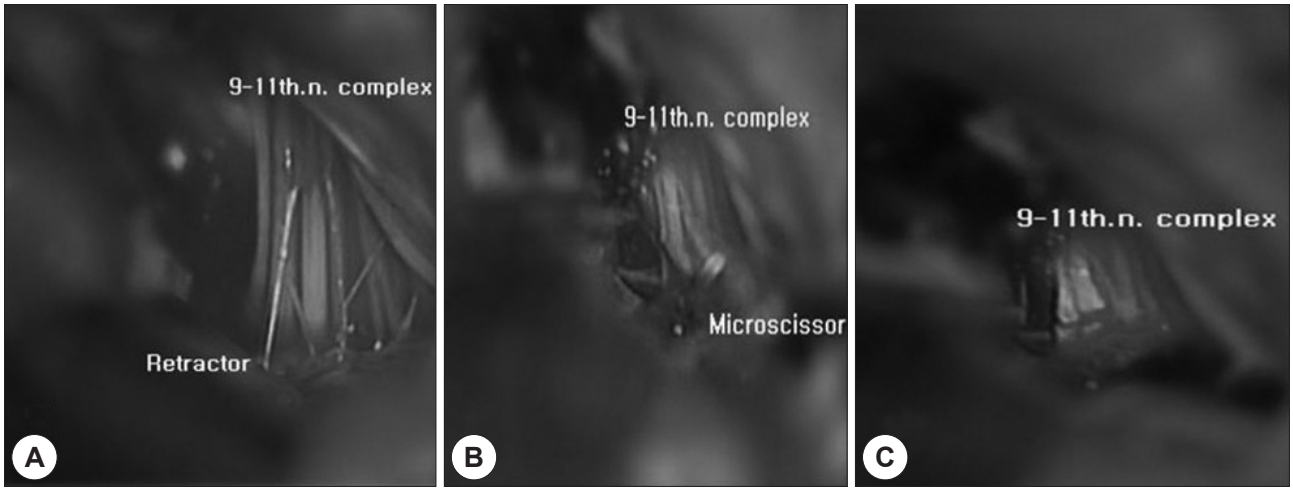

Fig. 3. We dissected the arachnoid membrane of the entire length of the 9–11th nerve complex to attempt to isolate the vascular compression around the 9–11th nerve complex (A and B). The glossopharyngeal nerve was sectioned completely, and the upper rootlet part of the vagus nerve was sectioned (C).

DISCUSSION

Microvascular decompression (MVD) is considered the most efficient treatment for cranial neuralgia with offending vessels. It was first reported by Gardner in 1962 and the vascular compression hypothesis was further developed and popularized in 1967 by Jannetta.[3] The rhizotomy of the cranial nerve was performed when there was no compression. Further current treatments include percutaneous balloon microcompression, radiofrequency thermorhizotomy, drug injection within the trigeminal cistern,[4] stereotactic gamma knife surgery, first reported by Leksell, and linear accelerator (Linac)-based devices, only available in a few centers. There are also therapies for trigeminal neuralgia (TN) and glossopharyngeal neuralgia.[5] Unfortunately, in our case, there was no vessel compressing the trigeminal and glossopharyngeal nerve ; therefore, a rhizotomy of the trigeminal and glossopharyngeal nerve was performed.

Because of the TN, reports in the article are mostly focused on TN with vascular compression. In 1989, Adams critically discussed the concept of microvascular compression.[6] Many theories have been suggested but none has been scientifically proven. It is not clear why some individuals with vascular compression of the trigeminal nerve are pain free. Hardy and Rhoton studied 50 trigeminal nerve roots in 25 cadavers : 60% of nerves had a vascular contact but none of these people had TN during life.[7] Root compression alone is therefore not a specific cause of TN.[6] In 2002, Devor, et al. postulated the 'ignition' hypothesis in which the primary effect of vascular compression induces a localized trigeminal ganglion (TRG) which leads to hyperexcitability.[8] This would give rise to pain paroxysms because of synchronized post-discharge activity originating at ectopic pacemaker sites in the root or TRG. It is possible that, similar to ectopic pacemakers, the compression site is mechanosensitive.[9]

Glossopharyngeal neuralgia can be idiopathic or secondary to other causes. It does not have a defined etiology, like trigeminal neuralgia. Secondary glossopharyngeal neuralgia can occur due to compression of the glossopharyngeal nerve by vascular structures, lesions, or intracranial tumors, Paget's disease, a lengthened styloid process, occipital-cervical malformations, oropharynx and tongue tumors, and inflammatory processes, like Sjogren's syndrome.[10] Combined trigeminal neuralgia and glossopharyngeal neuralgia is difficult to diagnose because there is no objective test. Therefore, clinical symptoms should be observed in detail.

In a series by Young, et al.[11] a partial sensory rhizotomy was performed instead of, or in addition to, microvascular decompression because it was not clear that significant vascular contact with the trigeminal nerve root was present, the surgeon was unable to completely separate the vessels from the neural structures or thought that it was unsafe to do so, or the patient had obtained poor results from a prior microvascular contact at the time of the reoperation. In this case, rhizotomy was performed because there was no offending vessel ; in another case, rhizotomy was performed due to the poor results of a prior MVD. Surgically, for the partial sensory rhizotomy, half of the cross-sectional area of the sensory root of the trigeminal nerve was bisected in its caudolateral aspect approximately 2 to 5mm from the pons.[11]

Gudmundsson, et al.[12] found that the relationships among the three trigeminal divisions remain constant from the ganglion to the pons, despite the fact that "immediately posterior to the ganglion, many anastomoses were observed between the fibers from each division." By following the three divisions posteriorly from the gasserian ganglion, they found that "the fibers from the third division remained ventrolateral throughout the interval from the ganglion to the pons, the first division dorsomedial, with second division fibers being in an intermediate position".[12] This anatomical conception of the three trigeminal sections is advocated by clinical data. In Adams, et al.[13] patients who underwent caudolateral nerve root section had impaired sensation in the third and second divisions of the trigeminal nerve postoperatively. Most experienced good outcomes.[13] We sectioned the sensory root of the trigeminal nerve caudolateral aspect 3 mm from the pons. In a series by Bederson, et al.[14] 56 cases of vascular contact without distortion and 30 cases without extrinsic compression were surgically treated with partial sensory rhizotomy. Most patients experienced complete pain resolution after the operation.

Glossopharyngeal neuralgia without offending vessels was treated with rhizotomy in these reports. In a series by Kandan, et al.[15] 6 patients exhibited no vascular compression. They sectioned the glossopharyngeal nerve and the first two rootlets of the vagus nerve. There were no complications such as hoarseness, dysphasia, loss of taste sensation or CSF leakage, and the pain was resolved after the rhizotomy.[15] In a series by Rey-Dios, et al.[16] they summarized studies involving rhizotomy of the glossopharyngeal nerve and upper rootlets of the vagus nerve for glossopharyngeal neuralgia. In that paper, rhizotomy for glossopharyngeal neuralgia produced good outcomes.[16]

Craniotomy and posterior fossa exploration has a mortality rate of 5% ; complications such as sensory and motor deficits, perioperative haematoma, meningitis, as well as swallowing and phonation abnormalities may occur.[17] With percutaneous radiofrequency rhizotomy, mortality is rare but complications can occur and usually consist of sensory deficits ; vasomotor abnormalities may occur during the procedure and promote abortion of the treatment in some patients.[1] The advantages of percutaneous surgery (radiofrequency rhizotomy, trigeminal tractotomy) are reduced mortality and that it is not necessity to admit the patient to the hospital for observation, which results in a simpler, safer, and cheaper procedure to treat trigeminal and glossopharyngeal neuralgia. However, these procedures usually present a higher recurrence rate when compared to that of MVD and rhizotomy of the trigeminal and glossopharyngeal nerve. Risks of decompression include death (for 1% of patients), intracranial hemorrhage, and morbidity, such as dysphagia or swallowing difficulties.[18] Since there was no compression vessel, we performed a rhizotomy of a quarter of the sensory part of the trigeminal nerve and simultaneously performed a rhizotomy of the glossopharyngeal nerve and the upper rootlet of the vagus nerve.

CONCLUSION

This case is a report of a rhizotomy for combined trigeminal and glossopharyngeal neuralgia without an offending vessel. Of the papers on cranial neuralgia in the literature, there were few reports on combined trigeminal and glossopharyngeal neuralgia without an offending vessel. Thus, it is difficult to determine the appropriate treatment. Based on the preoperative examinations, when an offending vessel was not found, we determined that rhizotomy was the proper treatment. Rhizotomy may therefore improve the likelihood of long-term pain control in patients diagnosed with combined trigeminal and glossopharyngeal neuralgia in the absence of an offending vessel.

REFERENCES

1. Rushton JG, Stevens JC, Miller RH: *Glossopharyngeal (vagoglossopharyngeal) neuralgia: a study of 217 cases. Archives of Neurology 38:201-205, 1981*
2. Jannetta PJ: *Neurovascular compression in cranial nerve and systemic disease. Annals of Surgery 192:518-525, 1980*
3. Gardner WJ: *Concerning the mechanism of trigeminal neuralgia and hemifacial spasm. Journal of Neurosurgery 19: 947-958, 1962.*
4. Hakanson S: *Trigeminal neuralgia treated by the injection of glycerol into the trigeminal cistern. Neurosurgery 9:638-646, 1981*
5. Leksell L: *Sterotaxic radiosurgery in trigeminal neuralgia. Acta Chirurgica Scandinavica 137:311-314, 1971*
6. Adams CB: *Microvascular compression: an alternative view and hypothesis. Journal of Neurosurgery 70:1-12, 1989*
7. Hardy DG, Rhoton AL Jr: *Microsurgical relationships of the superior cerebellar artery and the trigeminal nerve. Journal of Neurosurgery 49:669-678, 1978*
8. Devor M, Amir R, Rappaport ZH: *Pathophysiology of trigeminal neuralgia: the ignition hypothesis. The Clinical Journal of Pain 18: 4-13, 2002*
9. Chen Y, Devor M: *Ectopic mechanosensitivity in injured sensory axons arises from the site of spontaneous electrogenesis. European Journal of Pain 2:165-178, 1998*
10. Urban PP, Keilmann A, Teichmann EM, Hopf HC: *Sensory neuropathy of the trigeminal, glossopharyngeal, and vagal nerves in Sjogren's syndrome. Journal of the Neurological Sciences 186:59-63, 2001*
11. Young JN, Wilkins RH: *Partial sensory trigeminal rhizotomy at the pons for trigeminal neuralgia. Journal of Neurosurgery 79:680-687,*

1993

12. Gudmundsson K, Rhoton AL Jr, Rushton JG: *Detailed anatomy of the intracranial portion of the trigeminal nerve. Journal of Neurosurgery 35:592-600, 1971*
13. Adams CB, Kaye AH, Teddy PJ: *The treatment of trigeminal neuralgia by posterior fossa microsurgery. Journal of Neurology, Neurosurgery, and Psychiatry 45:1020-1026, 1982*
14. Bederson JB, Wilson CB: *Evaluation of microvascular decompression and partial sensory rhizotomy in 252 cases of trigeminal neuralgia. Journal of Neurosurgery 71: 359-367, 1989*
15. Kandan SR, Khan S, Jeyaretna DS, Lhatoo S, Patel NK, Coakham HB: *Neuralgia of the glossopharyngeal and vagal nerves: long-term outcome following surgical treatment and literature review. British Journal of Neurosurgery 24:441-446, 2010*
16. Rey-Dios R, Cohen-Gadol AA: *Current neurosurgical management of glossopharyngeal neuralgia and technical nuances for microvascular decompression surgery. Neurosurgical Focus 34:E8, 2013*
17. Teixeira MJ, de Siqueira SR, Bor-Seng-Shu E: *Glossopharyngeal neuralgia: neurosurgical treatment and differential diagnosis. Acta Neurochirurgica 150:471-475; discussion 475, 2008*
18. Taha JM, Tew JM Jr: *Long-term results of surgical treatment of idiopathic neuralgias of the glossopharyngeal and vagal nerves. Neurosurgery 36:926-930; discussion 930-921, 1995*