



Idiopathic Superior Laryngeal Neuralgia Cured by Microvascular Decompression

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Abstract

Background and importance: To date, very few SLN cases have been reported, and current treatments for SLN are mainly drugs, nerve block and nerve section. We encountered a case of Idiopathic Superior Laryngeal Neuralgia (ISLN) cured by Microvascular Decompression (MVD) when carbamazepine failed, and this is the first English report on an ISLN cured via MVD worldwide.

Clinical presentation: A 62-year-old female farmer was admitted due to a three year history of severe left throat lancinating. Carbamazepine at a dose of 450 mg per day incompletely alleviated symptoms. The patient underwent surgical MVD of the left vagus nerve and was pain free thereafter.

Conclusion: ISLN treated by MVD has not been previously reported in English literature. We recommend MVD as the first option for intractable ISLN.

Keywords: Idiopathic superior laryngeal neuralgia; Microvascular decompression; Vagus nerve

Abbreviations

ISLN: Idiopathic Superior Laryngeal Neuralgia; SSLN: Secondary Superior Laryngeal Neuralgia; MRI: Magnetic Resonance Imaging; MVD: Microvascular Decompression; PICA: Posterior Inferior Cerebellar Artery

Background and Importance

Superior Laryngeal Neuralgia (SLN) is a rare entity first described in 1900 by Avellis [1]. SLN is characterized by severe pain in the lateral aspect of the throat, the submaxillary region, and/or under the ear. It is a very rare form of neuralgia that is generally treated with carbamazepine [2,3], local nerve block [4,5], and nerve section. SLN is classified as Idiopathic Superior Laryngeal Neuralgia (ISLN) or Secondary Superior Laryngeal Neuralgia (SSLN). ISLNs cured *via* MVD haven't been reported in English literatures worldwide. Here, we present a case of ISLN cured *via* MVD and discusses its etiology, classification, and treatment.

Case Presentation

A 62-year-old female patient presented at our hospital on 22 March 2017 with a three year history of severe left throat lancinating pain. Each episode lasted several minutes and radiated to the external auditory canal without being induced by drinking or taking food. The patient underwent electronic laryngoscopy and cervical MRI in the E.N.T. clinic, and the results were normal. Cranial base MRI showed that the left Posterior Inferior Cerebellar Artery (PICA) formed a vascular loop and compressed the left posterior cranial nerves (Figure 1). The patient was diagnosed with ISLN and administered carbamazepine at a dose of 450 mg per day, which only alleviated her symptoms for two months and later failed. The patient requested further treatment and was admitted to our department. She had no positive neurological findings except a trigger point at the left superior area of the thyroid cartilage. We obtained informed consent from the patient and her family and chose to perform an MVD for the patient. A standard left retrosigmoid approach was performed. After general anesthesia and semipronation, step-by-step, the posterior cranial nerves were exposed clearly. As seen in the picture, the vagus nerve was compressed by the PICA loop (Figure 2); the vertebral artery was seen adjacent to the left vagus nerve (Figure 3); finally vagus nerve was well-decompressed (Figure 4). The patient's pain was cured without neurological deficits postoperatively, and she was discharged after five days. We have followed this patient for more than one and a half years, and her pain has not recurred.

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Received Date: 18 Oct 2019

Accepted Date: 08 Nov 2019

Published Date: 13 Nov 2019

Citation:

Yan K-K, Deng Z-Bi, Xu J, Wang L-Bi, He D-S. Idiopathic Superior Laryngeal Neuralgia Cured by Microvascular Decompression. *Clin Surg*. 2019; 4: 2652.

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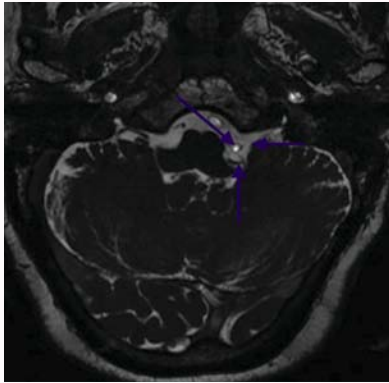


Figure 1: Constructive interference in steady state (CISS) reveals that the left posterior inferior cerebellar artery formed a vascular loop (vertical arrow) and compressed the left vagus nerve (horizontal arrow). The vertebral artery (diagonal arrow) was seen clearly.



Figure 2: Intraoperative picture of MVD showed that a vascular loop (vertical arrow) had compressed the left vagus nerve (horizontal arrow).

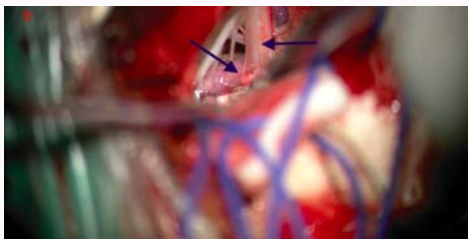


Figure 3: The vertebral artery (diagonal arrow) was seen adjacent to the left vagus nerve (horizontal arrow).

Discussion

Typical SLNs have been described as an idiopathic syndrome of paroxysmal pain generally confined to one side of the laryngeal region (left greater than right) and may radiate to the infra-auricular area. The act of swallowing may elicit pain, which leads the patient to avoid food and results in significant weight loss [6]. Pain worsened by swallowing is thought to be due to central causes. However, in our case the patient's pain was not elicited by swallowing. The superior laryngeal nerve arises from the vagus nerve in the area of the nodose ganglion and divides into external and internal branches. Pain can be triggered by pressure to the skin above and lateral to the thyroid cartilage where the internal branch of the superior laryngeal nerve pierces the thyrohyoid membrane. Anesthetizing the larynx with a topical anesthetic is an alternative method for confirming the diagnosis [7]. Etiology of the SLN remains unclear. SLN can be categorized into 2 broad categories: central and peripheral causes. The former, also named idiopathic SLN, is thought to be a compression of the upper fibers of the vagal nerve as they leave the brain stem and traverse the subarachnoid space to the jugular foramen, similar

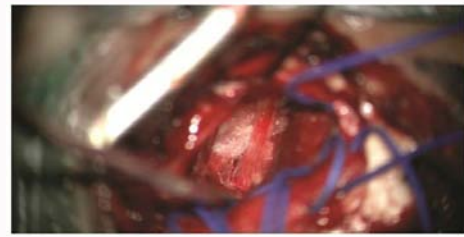


Figure 4: Intraoperative view of the MVD. The left vagus nerve was decompressed well with Teflon.

to other cranial nerve diseases. In the International Classification of Headache Disorders, 3rd edition (beta version), SLN is classified as glossopharyngeal neuralgia. The latter, also named secondary SLN is thought to follow causes, such as deviation of the hyoid bone [7], acute laryngitis [8], scarring from carotid artery surgery [9], trauma [10], microsurgery tonsillectomy [11], lateral pharyngeal diverticulum [12], etc. ISLN is also a diagnosis of exclusion. Thus, the differential diagnosis of ISLN includes such underlying structural conditions as inflammatory processes of the larynx and laryngeal neoplasms [5]. In our case, electronic laryngoscopy and cervical MRI ruled out these structural abnormalities. Other peripheral neuralgia diseases distinguished from SLN mainly depend on the location of pain and trigger points. For example, carotidynia resembling SLN is characterized by throbbing pain in the anterior cervical region that may be accompanied by migraine-like headache [13]. So far, treatments for ISLN are multitudinous and controversial, including oral drugs, such as lacosamide [14], carbamazepine [2,3,6], gabapentin [15], gamma knife radiosurgery [16], nerve block [4,5], and peripheral superior laryngeal neurotomy [17], and intracranial resection of the glossopharyngeal and upper vagal rootlets [18]. Early in 1995, Taha JM reported two cases of ISLN which were cured by peripheral superior laryngeal neurotomy but recurred 2 and 4 years postoperatively, respectively. These two patients were reported and cured via intracranial resection of the glossopharyngeal and upper vagal rootlets. In general, as we know, neuralgia, especially that resulting from central causes, would not be cured for a long time by oral medications, gamma knife radiosurgery or nerve block. Therefore, surgical options must be considered. We don't support peripheral superior laryngeal neurotomy as an option based on the general principles of cranial nerve diseases. We prefer to perform MVD than intracranial resection of the glossopharyngeal and upper vagal rootlets because the former is safer and minimally invasive unless performing MVD is difficult. We reviewed all the SCI and Chinese literatures associated with surgical therapies of ISLN and found only 6 articles (totaling 14 cases), and only an MVD treatment for ISLN was reported [19]. This article is the first English literature on using MVD for ISLN. We summarized the information for 15 ISLN patients. Because ISLNs are extremely rare and cases of invasive therapies are lacking, the abovementioned cases in Table 1 will still need abundant observation and follow-up. So far, which treatment method is better remains controversial, needing more cases to be verified. However, we propose that MVD may be the optimal option for well-diagnosed ISLN based on the general principles of cranial nerve diseases.

Conclusion

We performed a successful surgical treatment of ISLN *via* MVD. This is the first English report on ISLN treated by MVD worldwide.

We recommend MVD as the first option for intractable ISLN because it is safe and minimally invasive based on the general principles of cranial nerve diseases.

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