

## Excellent response to acupuncture treatment in a case of intractable glossopharyngeal neuralgia due to vascular tortuosity: a case report

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### Abstract

**Objective:** Glossopharyngeal neuralgia is a relatively rare neurological condition arising from abnormal irritation of the ninth cranial nerve. The first line of conventional treatment is pharmaceutical followed by surgical interventions in non-responsive cases. However, the efficacy and safety of usual therapeutic methods have limited the compliance of most affected patients. **Methods:** Here for the first time, we report an excellent response of a chronic case of glossopharyngeal neuralgia to traditional Chinese medicine, which is a relatively safe and not expensive therapeutic method. **Results:** Despite limited response to conventional medicine, our patient showed a fast and promising response to the combination of acupuncture and moxibustion with low doses of carbamazepine. **Conclusion:** The therapeutic protocol used for the treatment of this patient needs to be validated by further double-blinded case control studies in order to be compared to other available therapeutic methods.

**Key words:** Glossopharyngeal neuralgia, Facial pain, Acupuncture, Moxibustion

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**Abbreviations:** TCM, traditional Chinese medicine; CGRP, calcitonin gene-related peptide.

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## Background

The ninth cranial nerve or glossopharyngeal nerve consists of a mixture of cranial nerves which innervates oropharynx, Eustachian tube, and posterior third of the tongue as well as middle ear and mastoid process [1]. Abnormal irritation of this nerve causes glossopharyngeal neuralgia, which is a kind of neurogenic pain mainly in oropharynx from posterior area of throat, tonsillar fossa, base of tongue, ear canal to inferior of mandibular angle. It causes significant discomfort during activities requiring the movements of mandible such as speaking and swallowing [2]. Glossopharyngeal neuralgia happens in 0.2 to 0.4 cases per 100,000 people per year with no gender predilection. Its incidence is about 1/6 of trigeminal neuralgia, its twin sister [3]. Mainly affecting individuals older than 50, the incidence of glossopharyngeal neuralgia increases by age [4]. The pain is usually paroxysmal, unilateral, sharp, stabbing and severe shooting which lasts for seconds to minutes. It repeats after a certain, usually long, interval, but there are reports of repeated attacks in a day [2]. Some patients complain of constant symptoms lasting for days and months [5]. The pain is commonly triggered by swallowing, yawning, talking and coughing [6, 7].

In most cases, the pathophysiology behind the disease is the pressure effect of postero-inferior cerebellar artery [6]. The disorder may also be secondary to an insult to neighboring organs such as trauma, infection, radiation, neoplastic infiltration, surgical manipulations, vascular malformations, persistent hypoglossal artery, dissection of vertebral artery, multiple sclerosis, etc. [2]. The gold standard diagnostic method is Magnetic Resonance Imaging, which precisely visualizes the relationship between this nerve and the conflicting artery in the supraolivary fossa. At the same time, it can exclude most of other pathologies, which are listed as the secondary causes of glossopharyngeal neuralgia [8]. However, there are some idiopathic cases with no paraclinical findings. In such cases, there are usually some evidences of vascular decompression or central pontine dysfunction [2].

The treatment is mainly based on pain control with medical therapy using carbamazepine, gabapentin or pregabalin [2, 6] usually complemented by vitamin B12 and low doses of selective serotonin reuptake inhibitors [2]. Most patients also try NSAIDs or opioid pain killers as well as physical/psychological therapies to control the pain. However, the efficacy of these methods is controversial [9–11]. In non-responsive cases, some practitioners advise nerve block techniques using different combinations of anesthetic agents and other adjuvants. However, significant side effects of such procedures such as vocal cord paralysis

have limited their use [12]. Most often, after failure of less invasive methods, surgical microvascular decompression is considered [6]. Obviously, surgical procedures have the greater chances of trauma to the surrounding neuro-vasculature, hematoma, cerebrovascular accidents, vocal cords paralysis, dysphagia, tinnitus, vertigo, meningitis, etc. Furthermore, the recurrence rate after surgical intervention is high which has questioned the efficacy of these methods in controlling glossopharyngeal neuralgia [2, 7]. Recently, less invasive surgical methods such as CT-guided pulsed radiofrequency and Gamma-knife radiosurgery have been introduced with prominent success rates [13, 14]. However, high cost and limited regional availability may restrict their use in a global scale. Due to severity of symptoms, variable response rate to conventional treatments and limiting factors such as side effects, potential risks and financial matters, the need for an effective, available, sustainable and economic method of treatment is felt. Acupuncture can be a solution.

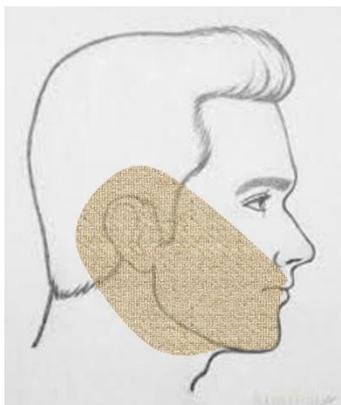
Acupuncture is one of the most ancient healing modalities dating back to 6000 BC. Although originated from China, it is practiced worldwide today [15]. In this practice, solid filiform needles are placed in specific points of body to promote the health and treat the illnesses [16]. Acupuncture is closely used together with moxibustion which involves the direct or indirect application of ignited mugwort (*Artemisia vulgaris*) on acupuncture points or other specific parts of the body to facilitate healing [17]. Despite lack of insurance coverage [18], it is shown that implication of acupuncture-based methods reduces the sum of medical expenditure [19, 20].

There are reports on administration of acupuncture for the treatment of head and neck problems such as facial pain [21] and paralysis [22] as well as cerebrovascular accidents [23]. In addition, the combination of Phenytoin and acupuncture has been shown to be beneficial in management of intractable facial pain and Bell's palsy [24]. However, there has not been any report on the effect of acupuncture-based interventions in the treatment of glossopharyngeal neuralgia with vascular pathophysiology, yet.

## Case presentation

The patient was a 53-year-old dentist complaining of pain at the root of his tongue since six years ago. By the time, the pain worsened and its area extended to the right side of mandible, buccal region and the right ear (Figure 1). At the first visit, he complained of persistent, deep, penetrating, sharp pain in his tongue and the right side of his face, which had affected his daily life, despite medical therapy. He encountered excessive ache while chewing and speaking. Therefore, he had limitation in solid food consumption and spoke

with closed mouth. He was on high doses of Carbamazepine, Gabapantin, and Opium at the time of the first visit.



**Figure 1** The affected area based on patient's complains

In physical examination, there was nothing significant except tender points in the right side of face mainly in mandibular angle, mastoid process and sub-zygomatic region. The oral cavity could not be examined since an intolerable pain was triggered by opening of the mouth. There was no visual or hearing loss or any other sensory problem. The temporomandibular joint was restricted in motion due to a significant spasm in the right masseter muscle.

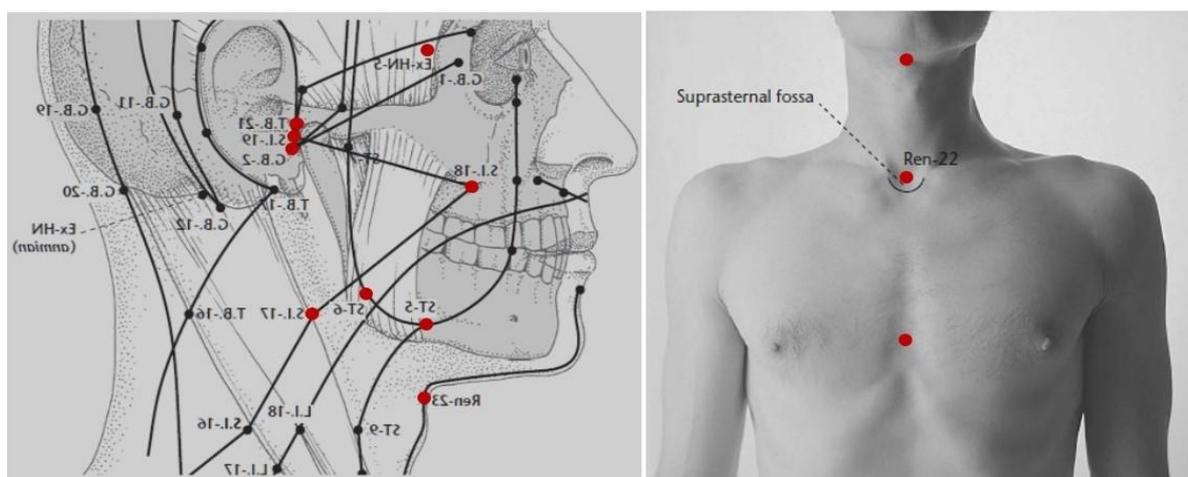
Magnetic Resonance Imaging with and without gadolinium contrast had shown the tortuosity of vertebrobasilar system with vascular compression of right vertebral artery over the right glossopharyngeal nerve without any abnormal enhancement after contrast injection. The Magnetic Resonance Angiogram and Magnetic Resonance venogram imaging were reported to be normal.

Since the start of his problem, he had tried different conventional and herbal medicines with minimal or no response. They included NSAIDs, anticonvulsants, antidepressants, Gabapentin, herbal opium based mixtures, and some other drugs, which he could not recall. After unsuccessful medical therapy, surgical consultation was done. Because of the potential risks associated with surgical intervention explained by surgeons, he refused to undergo any of the recommended surgical procedures.

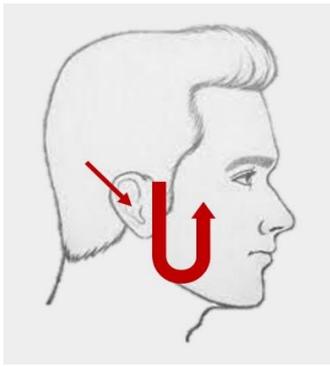
### Treatment protocol

The patient was evaluated based on Traditional Chinese Medicine (TCM). It was diagnosed as fixed or damp Bi-Syndrome in TCM. Hence, a combination of acupuncture and moxibustion was applied to treat the problem [25, 26]. Table 1 and Figure 2 demonstrate local and remote points, which were used in this case. The acupoints were selected based on the affected meridians and the territory of pain. In addition, since there was a significant spasm in affected muscles, Pi-Spleen Channel was used to regulate the muscle performance. The patient complained of severe mental disturbance due to chronic pain and unsuccessful therapeutic attempts. To tackle this, we tried to “Open the Four Gates” as well using LI4 and LV3 points bilaterally. After each acupuncture session, the pain territory was warmed up with moxa sticks (Figure 3).

The response to the treatment was evaluated at each session based on pain severity scale from 0 (no pain) to 10 (the extreme, unbearable pain). The pain was assessed in six categories consisted of: a) sharp penetrating pain in tongue; b) ear pain; c) pain during speaking; d) pain during drinking; e) pain on chewing; and f) pain during swallowing.



**Figure 2** Schematic location of acupoints used in the protocol based on the Standard Atlas of Acupuncture [38]



**Figure 3 Moxibustion protocol**

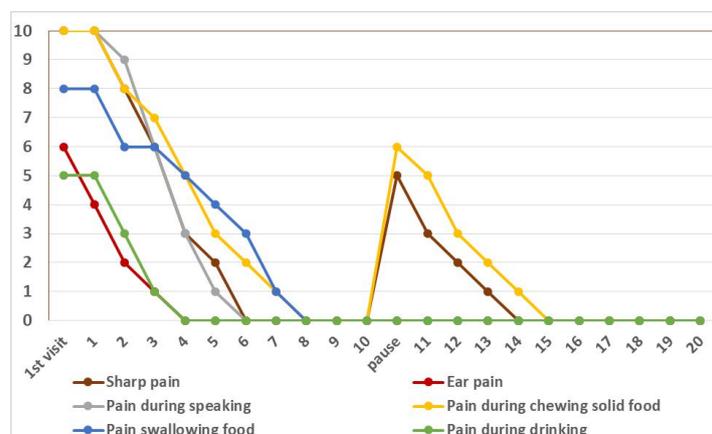
The protocol was administered every other day for the first week. After reaching a significant response, in the third session, it was continued twice a week. At the same time, the patient stopped using opium after the second session and started decreasing the

doses of pharmaceutical drugs. In the sixth session, he reported that he has discontinued the consumption of Gabapentin and he has decreased the Carbamazepine to half of his prescribed doses based on the consultation with his neurosurgeon. After 10 sessions, the patient was painless and treatment was paused for 2 weeks in order to prevent the point fatigue syndrome. During this treatment break, the patient experienced sharp pain in the right side of the face on the 10<sup>th</sup> day after some mental distress. It was a deep pain in the basis of his tongue and only triggered by chewing, but not speaking or swallowing. After that, the treatment was resumed twice weekly. Despite the complete response after another six sessions, the patient insisted on the continuation of the treatment weekly to prevent the recurrence of that exhausting pain.

**Table 1 The acupoints used in the protocol**

Channel	Point (s)	Position
Liver	3	Bilateral
Large Intestine	4	Bilateral
Spleen	6	Bilateral
Conception vessel (Ren Mai)	17, 22, 23	-
Stomach	4, 5*	Right
Small intestine	17*, 18*, 19	Right
Gall bladder	2	Right
Triple energizer	21	Right
Extraordinary head and neck	3	Right
Scalp	Sensory of face	Right

NOTE. \* points used for needle-moxibustion.



**Figure 4 Severity of pain reported by the patient in different sessions.** The session zero was the date of primary visit before the start of protocol. The pause was lasted for two weeks.

## Treatment outcome

Figure 4 shows the severity of pain reported by the patient in different sessions. As can be seen, after the second treatment session, the patient had no ache during drinking and his ear pain was completely vanished. The penetrating sharp pain in tongue and painful speaking were completely relieved after four treatment sessions. While, the pain during chewing and swallowing took another two sessions to disappear. After ten treatment sessions, the protocol was paused for two weeks. He was symptom free for 10 days despite having no acupuncture sessions and decreasing the medicines used. Then, penetrating pain in tongue and pain during chewing solid food recurred. However, its severity was much less than the pain he suffered before the start of acupuncture sessions (Figure 4). Following that, another course of acupuncture was started for ten more sessions. It took only four more sessions to reach a complete response. However, the patient insisted on continuing the protocol weekly to prevent the recurrence of symptoms.

In order to see the effect of acupuncture treatment on pathophysiology of the disease, we requested him to repeat the magnetic resonance studies. However, the patient did not accept to undergo imaging procedures after treatment due to risks associated with covid-19 pandemic.

## Discussion and conclusion

Glossopharyngeal neuralgia usually comes under either fixed or aching Bi-Syndrome in Chinese Medicine. The word 'Bi' in Chinese means an obstruction. It is believed that the pain in patients diagnosed as Bi-syndrome comes from an obstruction in Qi and blood movement in the affected areas [25, 26]. Bi-syndrome is usually caused by invasion of external pathogens such as wind, cold or damp which can interfere with the normal movement of Qi and Blood in meridians and collaterals. If the symptoms are caused by invasion of pathologic wind, it will be wondering Bi-syndrome. Similarly, Aching, Fixed and Febrile Bi-syndromes are due to cold, damp and heat pathogens, respectively. Many rheumatologic disorders and neuralgias are categorized under this entity in Traditional Chinese Medicine [27, 28]. Based on TCM protocols, cold and damp Bi-syndrome problem can be treated by combination of acupuncture and moxibustion [26, 27].

The efficacy of acupuncture to relieve facial pain can be discussed via several mechanisms. They include its analgesic effects, anti-inflammatory mechanisms, improvement of local blood circulation, and the effect of this therapeutic method on the pathologic factors triggering the pain.

By insertion of acupuncture needles in the skin, a local vasodilation is usually observed characterized by circular redness and induration around the needles. It is suggested that Calcitonin Gene-Related Peptide (CGRP) released from A $\delta$  or C fibers is responsible for such temporary reaction [29]. CGRP is a bidirectional neuropeptide with anti-inflammatory effects in low doses and pro-inflammatory properties in higher doses. Its role in migraine headache and trigeminal neuralgia has been widely investigated [30–32]. Some contributions to CGRP mechanism can be referred to histamine release from local mast cells and its intrinsic inhibitor, nitric oxide [27]. Both of these molecules have been studied to be affected by acupuncture stimulation [33, 34]. In addition, there are studies proposing some acupuncture induced analgesia through the activation of a list of neuropeptides including opioid peptides, adenosine, anandamide, and norepinephrine [35]. Substance P, another vasoactive neuropeptide with analgesic effects might be also considered as a potential biomolecule involved in the analgesic action of acupuncture [29, 32]. Since there are limited publications on glossopharyngeal neuralgia, none of these biomolecular mechanisms can be suggested for the observed effect of acupuncture by certainty. Further studies are recommended for better understanding of the mechanism of action of acupuncture on glossopharyngeal neuralgia.

There are several available reports on effectiveness of acupuncture based methods in many causes of headache such as migraine, trigeminal neuralgia and maxillary pain [36, 37]. However, there is no publication on the efficacy of these methods on glossopharyngeal neuralgia. It can be due to low prevalence of this problem in the community. Here for the first time we reported the excellent response of intractable glossopharyngeal neuralgia due to vascular pressure effect to acupuncture and moxibustion. Such efficacy needs to be evaluated by other independent researchers and in case-control studies in order to be implemented in the routine therapeutic protocols. Additionally, longer follow-ups will be helpful to see how long the effects last after discontinuing the treatment [36].

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