ScientificScholar®
Knowledge is power
Publisher of Scientific Journals

www.surgicalneurologyint.com

Surgical Neurology International

Editor-in-Chief: Nancy E. Epstein, MD, Professor of Clinical Neurosurgery, School of Medicine, State U. of NY at Stony Brook.

SNI: Unique Case Observations

Editor
S. Ather Enam, MD, PhD
Aga Khan University, Karachi, Sindh, Pakistan

SNI
Open Access

Case Report

Concurrent glossopharyngeal neuralgia and oromandibular dystonia resolved after microvascular decompression of the trigeminal and glossopharyngeal nerve: A rare presentation

Wafa Faisal Aldhafeeri[1], Rahaf Farhan Alanazi[2,3], Jamal Abdullah[1], Abdullrahman Nazer[1]

[1]Department of Neurosurgery, Prince Sultan Military Medical City, [2]College of Medicine, King Saud bin Abdulaziz University for Health Sciences, [3]King Abdullah International Medical Research Center, Riyadh, Saudi Arabia.

E-mail: *Wafa Faisal Aldhafeeri - wfa.1995@gmail.com; Rahaf Farhan Alanazi - alanazirfd@gmail.com; Jamal Abdullah - drjamal.abdullah@gmail.com; Abdullrahman Nazer - abnazer@yahoo.com

***Corresponding author:**
Wafa Faisal Aldhafeeri,
Department of Neurosurgery,
Prince Sultan Military Medical City, Riyadh, Saudi Arabia.

wfa.1995@gmail.com

Received: 31 July 2023
Accepted: 22 March 2024
Published: 19 April 2024

DOI
10.25259/SNI_642_2023

Quick Response Code:

ABSTRACT

Background: This type of pain syndrome occurs suddenly and briefly, beginning unilaterally from one side of the face. Modestly stimulating speech can provoke it, affecting the ear, tongue, throat, and jaw angle. Interestingly, it is the sensory distribution of the auricular and the pharyngeal branches of the cranial nerves IX and X. People have not had a confirmed case of glossopharyngeal neuralgia (GPN), along with oromandibular dystonia (OMD). Nevertheless, usually in the medical literature, this case report supplies information about a patient who has concurrent GPN and OMD.

Case Description: A 36-year-old male patient presented with a history of sudden onset of increasing electric pains, which were centered in the middle of the forehead to the depth of the throat and accompanied by uncontrolled movements, repetitive tongue protrusions, jaw movements, and recurrent pervasive gagging reflexes. Magnetic resonance imaging showed that a vascular loop of the superior cerebellar and anterior inferior cerebellar artery on the left side had crossed over and compressed those nerves. Decompression surgery in the left glossopharyngeal and trigeminal nerves cured all the symptoms.

Conclusion: The simultaneous occurrence of GPN and OMD is rare, complex, and challenging from the clinician's viewpoint in the management of similar but different pathologies. A detailed history was taken, and a radiological investigation was called to devise a management plan in the context of understanding the pathology of both disorders.

Keywords: Glossopharyngeal neuralgia (GN), Microvascular decompression (MVD), Oromandibular dystonia (OD)

INTRODUCTION

Glossopharyngeal neuralgia (GPN) is an episodic syndrome estimated to account for an incidence of 0.2–1.3% of all cases of cranial neuralgia.[4] The typical characteristics include very rapid unilateral pain of concise duration in the distribution of the auricular and pharyngeal branches of the glossopharyngeal (IX) and vagus (X) cranial nerves.[8] Oromandibular dystonia

This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-Share Alike 4.0 License, which allows others to remix, transform, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.
©2024 Published by Scientific Scholar on behalf of Surgical Neurology International

(OMD) is a rare focal neurological disorder characterized by involuntary movements at the jaw-facial musculature and the tongue.[11] The current male presented with GPN associated with OMD, improving significantly post-microvascular decompression of the glossopharyngeal and trigeminal nerve. We now present an extensive literature review on the disorders and their management.

CASE PRESENTATION

History for a 36-year-old previously medically free man was presented in the clinic with a long-standing history of abnormal facial movements and facial pain. He was labeled rediagnosed by many neurologists as trigeminal neuralgia (TGN) and underwent a trial line of medical therapy without a pronounced response. A case is presented with sudden electric pain in the middle of the forehead accompanied by a feeling of this kind down in the throat followed by an involuntary uncontrolled movement as repetitive gag reflexes with tongue protrusion and jaw movements lasting only a second. These symptoms are usually precipitated by eating and drinking and worsen at nighttime. The clinical picture of the case showed a string susceptibility of co-occurrence of OMD and GPN. Accordingly, the treatment plan included adding the Movement Disorder Neurologist, and the patient had been initiated on clonazepam. During his follow-up at the clinic, his involuntary movements came under control; however, due to the tapering down of other medicines, the electrical pain component overrode it.

The radiological images in the multiple magnetic resonance imaging, magnetic resonance angiography, and fast imaging employing steady-state acquisition images indicated that there existed a constant offending vascular loop of the superior cerebellar artery as well as that of the anterior inferior cerebellar artery, crossing over to cause compression of the glossopharyngeal and the trigeminal nerve on the left side of the individual [Figure 1].

Microvascular decompression over the left side of the glossopharyngeal nerve, together with the trigeminal nerve, was done. Intraoperative findings were offending vessels, which were isolated and dissected away from the nerves by Teflon material application [Figure 2].

The patient gradually improved postoperatively till the complete resolution of his symptoms after one year. The patient was following up regularly, with a trial of weaning his current medicines for about a year without any relapse.

DISCUSSION

Trigeminal and glossopharngeal neurolgia accounts for only 5% of all GPN patients.[5,7] Diagnosis – GPN should take place in the case of manifestations of neuralgic pain, pharyngeal or

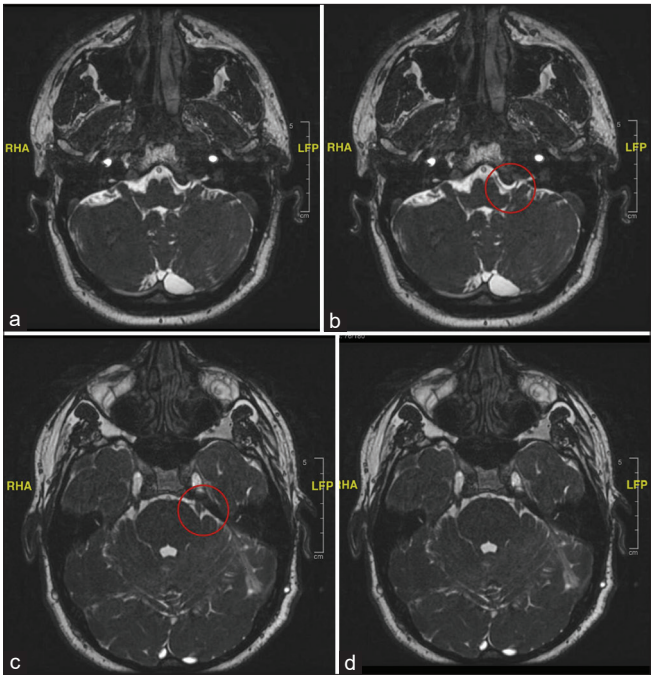

Figure 1: (a and b) Magnetic resonance imaging FIESTA sequence: at the level of glossopharyngeal nerve on the left side consistent with an offending loop by both SCA and AICA causing compression at root entry zone and crossing over of glossopharyngeal nerve. Here, the red circle depicts the above-mentioned findings. (c and d) Magnetic resonance imaging FIESTA sequence: at the level of Meckel cave consistent with an offending loop by both SCA and AICA causing compression at root entry zone and crossing over of trigeminal nerve. Here, the red circle depicts the above-mentioned findings. FIESTA: Fast imaging employing steady-state acquisition, SCA: Superior cerebellar artery, AICA: Anterior inferior cerebellar artery.

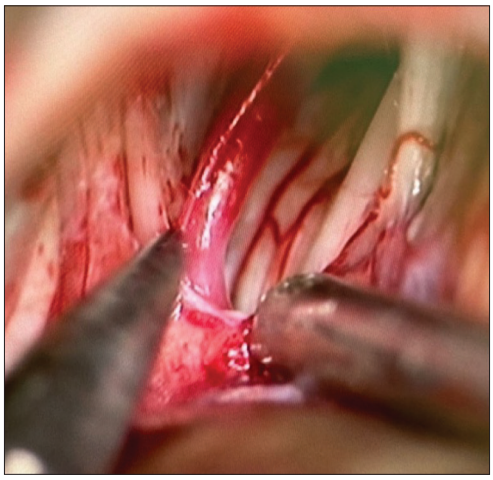

Figure 2: Intraoperative imaging through left retro sigmoid approach for microvascular decompression of an offending vascular loop, here depicted an offending vascular loop with compression around the entry point of the trigeminal nerve.

otalgic, with differential diagnosis of both GPN and TGN pain. The explanation of coexistence lies in the proximity of the area of the brainstem. Peripheral connections and central convergence of the nerves also contribute.[1] However, no prior reports were found regarding cranial nerve neuralgia with other motor disorders of the oromandibular region. In general, the planning of treatment is customized by the backdrop of each case and guided by a multidisciplinary approach.

Of course, it might be acceptable to approach microvascular decompression for GPN as a second-line treatment when standard medical treatment, for some reason, did not give the expected results if the pain relief was not achieved or the side effects of the drugs became unbearable. Appropriate specialized magnetic resonance imaging investigations are indispensable to visualize neurovascular compression in the root entry zone of the cranial nerve. Experienced neurologists request these and include 3D-constructive interference in the steady state.

Gaul *et al*. reported pain resolution in 16 of the 18 patients with GPN following MVD.[2,8] In other studies, reported pain-free rates were 76% of the patients with GPN, with an additional 16% of patients who improved.[9] Most patients with GPN remain pain-free at ten years postoperatively.[5,9]

At present, the pathophysiology of OMD remains to be unraveled. Trauma peripherally is considered one of the predisposing factors for several neurological disorders, including OMD.[6,10,11] The high possibility of either a causal relationship between the onset of OMD and neurovascular compression or the predisposing factors in the development of OMD was reported.[3] Yet, the correlative facts have not been pinpointed, and the requirement is open for more studies in this particular direction. The current case represents a patient with GPN and secondary OMD. Surgery with decompression of the 5th and 9th cranial nerves was performed in the neurosurgery department. It is necessary to mention that the treatment of GPN and OMD is complex and has a very diverse character, corresponding to the disease's severity and reason for its induction. They are approached similarly but naturally require surgical interventions in non classical treatment cases to be successful.

CONCLUSION

In conclusion, the simultaneous occurrence of Glossopharyngeal Neuralgia (GPN) and Oromandibular Dystonia (OMD) presents a rare and complex challenge for clinicians. This case study highlights the importance of a thorough history and radiological investigation in understanding and managing these disorders. Microvascular decompression surgery effectively relieved the symptoms in this patient, underscoring the significance of a multidisciplinary approach in treatment planning. While GPN typically responds well to microvascular decompression, the pathophysiology of OMD remains less understood, necessitating further research. Overall, this case emphasizes the need for tailored treatment strategies guided by the individual characteristics and underlying pathology of each disorder.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

REFERENCES

1. Bruyn GW. Glossopharyngeal neuralgia. Cephalalgia 1983;3:143-57.
2. Gaul C, Hastreiter P, Duncker A, Naraghi R. Diagnosis and neurosurgical treatment of glossopharyngeal neuralgia: Clinical findings and 3-D visualization of neurovascular compression in 19 consecutive patients. J Headache Pain 2011;12:527-34.
3. Jankovic J, Van Der Linden C. Dystonia and tremor induced by peripheral trauma: predisposing factors. J Neurol Neurosurg Psychiatry 1988;51:1512-9.
4. Katusic S, Willaims DB, Beard CM, Bergstralh EJ, Kurland LT. Epidemiology and clinical features of idiopathic trigeminal neuralgia and glossopharyngeal neuralgia: similarities and differences, Rochester, Minnesota, 1945-1984. Neuroepidemiology 1991;10:276-81.
5. Patel A, Kassam A, Horowitz M, Chang YF, Barrow DL, Sindou M, *et al*. Microvascular decompression in the management of glossopharyngeal neuralgia: Analysis of 217 cases. Neurosurgery 2002;50:705-11.

6. Raoofi S, Khorshidi H, Najafi M. Etiology, diagnosis and management of oromandibular dystonia: An update for stomatologists. J Dent 2017;18:73-81.
7. Rozen TD. Trigeminal neuralgia and glossopharyngeal neuralgia. Neurol Clin 2004;22:185-206.
8. Rushton JG, Stevens JC, Miller RH. Glossopharyngeal (vagoglossopharyngeal) neuralgia: A study of 217 cases. Arch Neurol 1981;38:201-5.
9. Sampson JH, Grossi PM, Asaoka K, Fukushima T, Casey KF, Jannetta PJ, *et al*. Microvascular decompression for glossopharyngeal neuralgia: Long-term effectiveness and complication avoidance. Neurosurgery 2004;54:884-90.
10. Sankhla C, Lai EC, Jankovic J. Peripherally induced oromandibular dystonia. J Neurol Neurosurg Psychiatry 1998;65:722-8.
11. Skármeta NP, Espinoza-Mellado P, PedroChana, Skármeta NP, Espinoza-Mellado P, Chana P. Orofacial dystonia and other oromandibular movement disorders. In: Dystonia: Different prospects. London: Intechopen; 2018.

How to cite this article: Aldhafeeri WF, Alanazi RF, Abdullah J, Nazer A. Concurrent glossopharyngeal neuralgia and oromandibular dystonia resolved after microvascular decompression of the trigeminal and glossopharyngeal nerve: A rare presentation. Surg Neurol Int. 2024;15:132. doi: 10.25259/SNI_642_2023

Disclaimer

The views and opinions expressed in this article are those of the authors and do not necessarily reflect the official policy or position of the Journal or its management. The information contained in this article should not be considered to be medical advice; patients should consult their own physicians for advice as to their specific medical needs.