PAIN MEDICINE CASE REPORTS

ISSN 2768-5152

Volume 7, Number 3, pp. 129-134

Recrudescing Trigeminal Neuralgia: A Chronicle of Orofacial Pain Before and After Cervical Cord Compression: A Case Report

Peter D. Vu, MD[1,2], Jeremiah F. Ling, BS[3], Michael V. Nguyen, MD[1,2], and Nikola Dragojlovic, DO[1,2]

Background: Classic trigeminal neuralgia is a clinical condition with pathognomonic acute and sporadic episodes of pain. Atypical trigeminal neuralgia manifests more dull, constant, and chronic with occasional acute burning or stabbing pain as experienced in classic trigeminal neuralgia. There have been reports of trigeminal neuralgia developing after cervical insults, positing a cervical source of orofacial pain.

Case Report: We report a patient with chronic, bilateral atypical trigeminal neuralgia symptoms due to a cervical cavernous malformation who underwent a C3-C6 laminectomy with cavernous malformation resection and had subsequent seroma-induced bilateral classic trigeminal neuralgia. Therapist-led cervical exercises improved his symptoms. His quality of life and function independence improved and his medications were titrated down.

Conclusions: In conclusion, trigeminal neuralgia does not always present as classic paroxysmal, electrical pain and can occur from a cervical insult distal to the C2 spinal level.

Key words: Trigeminal neuralgia, cervical cord compression, case report

BACKGROUND

Trigeminal neuralgia, also known as tic douloureux, is a clinical condition triggered by innocuous stimuli presenting as an acute, intense, paroxysmal electric shock along the territorial divisions of the trigeminal nerve (1,2). Although often characterized by classic paroxysmal facial pain, 24%-49% of patients with trigeminal neuralgia also report continuous, aching, chronic pain (3). The most common etiology of trigeminal neuralgia involves compression of the trigeminal nerve and may be secondary to demyelination at the site of compression (1,3). There have been reports of trigeminal neuralgia developing after cervical insults, however, positing a cervical source of orofacial pain (4-8). We report a patient with chronic bilateral trigeminal neuralgia symptoms due to a cervical cavernous malformation who underwent a C3-C6 laminectomy with cavernous malformation resection and the subsequent sequelae.

CASE PRESENTATION

The patient provided consent and approval to report this case for educational purposes. IRB approval was not required with deidentification and anonymization of identifiable medical information, per our institution's IRB protocol.

We report a 62-year-old man who presented to inpatient rehabilitation after undergoing an elective C3-C6 laminectomy and cavernous malformation resection

From: [1]McGovern Medical School at The University of Texas Health Science Center at Houston, Houston, TX; [2]TIRR Memorial Hermann, Houston, TX; [3]Texas A&M University Health Science Center College of Medicine, Bryan, TX

Corresponding Author: Peter D. Vu, MD, E-mail: peter.d.vu@uth.tmc.edu
Disclaimer: There was no external funding in the preparation of this manuscript.
Conflict of interest: Each author certifies that he or she, or a member of his or her immediate family, has no commercial association (i.e., consultancies, stock ownership, equity interest, patent/licensing arrangements, etc.) that might pose a conflict of interest in connection with the submitted manuscript.
Patient consent for publication: The patient provided informed consent to participate in this case report.
Authors adhere to the CARE Guidelines for writing case reports and have provided the CARE Checklist to the journal editor.
Accepted: 2023-02-07, Published: 2023-05-31

(Fig. 1). Prior to this procedure, the patient's medical history was significant for depression, anxiety, and a 6-year history of worsening chronic, dull pain in his shoulders, posterior neck, and bilateral face. For the past year, he reported an increasing frequency of dizziness, headaches, and acute electrical-numbing facial pain. His neurologist initially diagnosed him with Meniere disease, but several months later he started having progressive left hemiparesis and left arm neuropathic pain and spasms, prompting further workup with imaging. His brain magnetic resonance imaging was negative; his cervical spine magnetic resonance imaging, however, revealed a left C5 1.2 cm tapered intramedullary cavernous malformation with narrowing of the spinal canal from C3 through C6 (Figs. 2A and 2B). He subsequently underwent a laminectomy and cavernous malformation resection with resolution of his facial symptoms and improvement of his shoulder and neck pain. He was then discharged to inpatient rehabilitation.

At inpatient rehabilitation, the patient was started on celecoxib, hydrocodone-acetaminophen, and lidocaine patches for postoperative surgical pain resulting in good analgesic effects. A day after presenting to inpatient rehabilitation, however, he began experiencing facial fullness and acute pain exacerbated with light touch, prompting additional imaging. Computed tomography imaging of the sinus was negative for sinus disease, acute fractures, or apparent trigeminal nerve compression. However, a computed tomography image of the cervical spine revealed a new interval dorsal seroma at the C3 level, impinging on the spinal canal (Fig. 3).

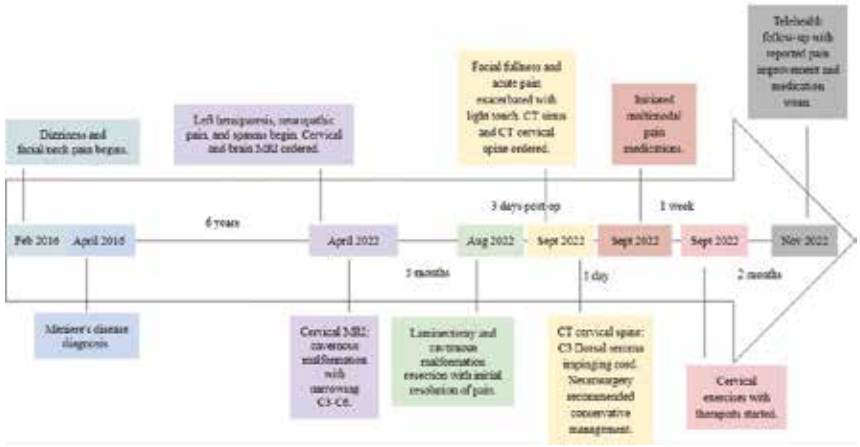

Fig. 1. Timeline of the patient's clinical presentation.

The neurosurgery department

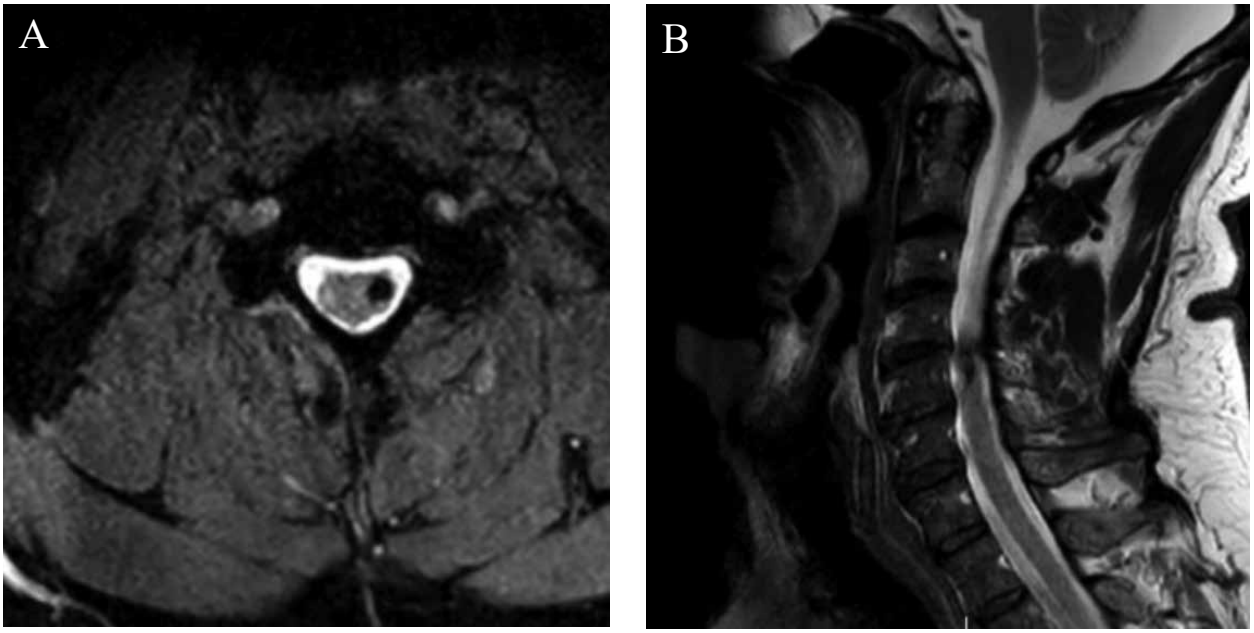

Fig. 2. Left cervical spinal cord C5 level tapered intramedullary lesion with hemosiderin representing a cavernous malformation. (A. sagittal plane; B. axial plane).

was made aware of the new imaging findings; conservative pain management was recommended. The patient agreed and was managed with titratable doses of gabapentin, duloxetine, and baclofen, resulting in only a marginal improvement in his facial pain. During this time the patient continued participating in physical therapy. He reported a substantial improvement in his facial pain with therapist-led posturing, isometric strengthening, prone and supine head lifts, neck rotations, and scapular retractions. With further inpatient therapy, the patient's pain was alleviated, his gabapentin was reduced, and his functional independence and quality of life improved. On telehealth follow-up 2 months after his discharge, the patient reported near resolution of his facial and posterior neck pain. He has begun weaning from baclofen.

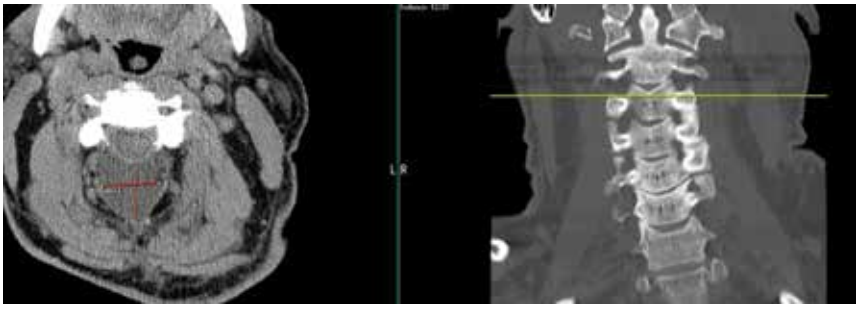

Fig. 3. Postoperative seroma dorsal thecal sac at C3.

DISCUSSION

Although cavernous malformation-induced unilateral trigeminal neuralgia is rare, with less than 20 reported cases (6-8), the presentation of bilateral atypical trigeminal neuralgia followed by bilateral seroma-induced trigeminal neuralgia is unique. Several classifications of trigeminal neuralgia exist: classic, atypical, and idiopathic (1,2). Classic presents pathognomonic acute and sporadic episodes of pain lasting one to 2 minutes. The pathophysiology is hypothesized to be compression of the sensory portion of the trigeminal nerve at the root entry of the pons by the superior cerebellar artery (1). Atypical manifestation may present with more dull, constant, and chronic pain with occasional acute burning or stabbing pain as experienced in classic (2). These attacks are often less severe than classic trigeminal neuralgia and are attributable to neurological causes, such as multiple sclerosis, malignancy, or mass effect causing neurovascular compression (1,2,9). Idiopathic trigeminal neuralgia, defined as evident facial pain with no discernible neurovascular or secondary etiology on imaging or neurophysiologic tests, is more uncommon than classic or atypical (1,2).

The initial resolution of the patient's atypical trigeminal neuralgia post cavernous malformation-resection followed by seroma-induced trigeminal neuralgia suggests a cervical etiology (4,10). Anatomical studies have shown the trigeminal nerve brainstem nuclei converging between the dorsal horn of the medulla and the upper levels of the cervical spine, projecting the spinal trigeminal nucleus to the C2 spinal level to merge with gray matter in this region (11-13). The patient's posterior cervical pain also postulates the involvement of the greater occipital nerve and third occipital nerve, which also resides in this region and provides cutaneous skin innervation along the occiput at C2 and C3 (14-16). The improvement of the patient's symptoms with cervical exercises, including semispinalis capitis usage, further supports the involvement of the third occipital nerve, which innervates that muscle (17). Electrophysiologic testing of cervical roots and the trigeminal nerve has reproduced both cranial and cervical symptoms respectively (10,18,19), further supporting the notion of convergence between trigeminal and cervical pain receptors.

Our discussions with the neurosurgery department and physical therapists provided a plethora of management options for this patient. Medical management with anticonvulsant agents carbamazepine, or oxcarbazepine is considered the first-line treatment for classic paroxysmal trigeminal neuralgia, with significant resolution of pain in approximately 90% of patients (1,2,9,20). Second-line medications, such as gabapentin, baclofen, and antidepressant agents such as nortriptyline and venlafaxine, have shown effectiveness and safety in treating continuous trigeminal neuralgia (1,2,9,20).

We titrated gabapentin to target the patient's trigeminal neuralgia and neuropathic pain, with minimal resolution of his symptoms. Duloxetine was utilized primarily for his anxiety and depression, with the additional benefit of treating his neuropathic pain. Baclofen helped manage his spasms, with secondary benefits targeting his other symptoms. We continued his celecoxib, lidocaine patches, and hydrocodone-acetaminophen for breakthrough pain.

Postsurgery, the care team discussed possible intervention treatments to treat his trigeminal neuralgia.

Percutaneous interventions, such as radiofrequency ablation, glycerol rhizotomy, and balloon compression have been standard procedures to treat classic trigeminal neuralgia. Each utilizes needles to target the trigeminal ganglion and its exiting branches at the foramen ovale to introduce heat, chemical, or mechanical damage, respectively (21). Gamma knife radiosurgery has gained traction as a nonincisional intervention, effectively delivering beams of highly focused radiation to the target tissue (22). Surgically, microvascular decompression is becoming the prominent intervention with a high response and high relief duration with minimal side effects compared to the aforementioned interventions (21,23). None of these procedures were considered for our patient, however, especially because of the known seroma visualized on imaging. In many cases, seromas are self-limiting and resorbed over weeks; surgical revisions are only considered when functional impairment, repetitive seroma formations, and wound infections are prevalent (24).

Literature on the role of physiotherapy with trigeminal neuralgia is limited and inconclusive (25), despite empirical evidence for it (26-31). The North American Spine Society claims cervical exercises can keep patients active and reduce cervical-induced pain (32). In the case of trigeminal neuralgia, 2 sources of impairment exist: 1) organic pathology and 2) deconditioning and muscle inactivity. Deconditioning decreases muscle strength and increases joint stiffness and postural strain, especially in patients with trigeminal neuralgia who attempt to minimize pain-induced movements (32).

Our patient's therapist-led exercises focused on head and cervical posturing, isometric strengthening, prone and supine head lifts, neck rotations, and scapular retractions to strengthen and stabilize cervical-induced pain. The patient's substantial improvement in pain after these therapies supports the possibility of cervical seroma-induced trigeminal neuralgia, similar in presentation to his chronic, cavernous malformation-induced symptoms. Considering the anatomical discussion above, the improvement of his symptoms predicates a cervical-trigeminal relationship that should further be explored. Several studies have shown the effectiveness of physiotherapy on seroma management (33-35). It is plausible that active mobility disrupts the fluid buildup and pressure the seroma places on the spinal trigeminal nucleus extending into C2, promoting faster resorption and improved symptoms.

Our main limitation for this case is a lack of consistent follow-up for this patient, since his primary residence is in another city from ours. We addressed that deficiency with virtual visits, where he last reported continued improvement of his pain and further down-titration of gabapentin. He has also been weaning off baclofen as well. He follows up with another physiatrist for continued rehabilitation needs. He is still participating in cervical exercises. Another limitation is the lack of generality because of the small sample size; however, the purpose of case reports is to highlight novel presentations and ideas that could result in increasing studies of similar presentations as an educational tool.

CONCLUSION

Although cavernous malformation-induced unilateral trigeminal neuralgia is rare, the presentation of bilateral atypical trigeminal neuralgia followed by seroma-induced bilateral classic trigeminal neuralgia is unique. Additionally, our case is unusual in that cases discussed above report proximal central cavernous malformation lesions while our patient's cavernous malformation lesion was more distal. Thus, we offer insight into a novel presentation of trigeminal neuralgia post-cervical laminectomy and cavernous malformation resection. A review of clinical presentations of trigeminal neuralgia and discussion of the anatomy has focused on the relationship the trigeminal nerve has with the cervical nerves, third occipital nerve, and greater occipital nerve. Our discussions with the neurosurgery department and physical therapists provided a plethora of management options, of which cervical therapies provided the most relief. Thus, in the case of facial pain, trigeminal neuralgia does not always present as classic paroxysmal, electrical pain and can occur from a cervical insult distal to the C2 spinal level. We recommend considering abnormal presentations of trigeminal neuralgia and considering management beyond pharmacology and surgery options.

Patient Perspective

The following was written by the patient in this case report:

Having the neck procedure saved my life and gave me the chance to recover it. Being in inpatient rehab has been a hard, long journey. I was glad that my facial pain improved initially after the surgery, however, when it came back I was distraught. My doctors tried a lot of pain medications for my neuropathy and my facial pain. The gabapentin, duloxetine, and baclofen were not that helpful. The Norco (hydrocodone-acetaminophen) was

able to dampen some pain. My biggest relief came with my physical therapists and their exercises and techniques to stretch my neck and shoulders. We did that before and after every session, and I did those exercises as many times as I could during my breaks. That has provided me the biggest relief in my facial pain and neuropathy. I am glad it allowed us to slowly wean some of the other medications.

Author Contributions

PV: Wrote the manuscript with JL with support from MN and ND.

JL: Wrote the manuscript with PV with support from MN and ND.

MN: Provided discussion and edits for the manuscript. Provided supervision with the patient's care.

ND: Conceived of the presented idea. Provided discussion and edits for the manuscript. Provided supervision with the patient's care.

All authors participated in medical care for the patient, discussed and interpreted the findings, and contributed to the final manuscript.

REFERENCES

1. Cruccu G, Di Stefano G, Truini A. Trigeminal neuralgia. *N Engl J Med* 2020; 383:754-762.
2. Jones M, Urits I, Ehrhardt K, et. al. A comprehensive review of trigeminal neuralgia. *Curr Pain Headache Rep* 2019; 23:74.
3. Di Stefano G, Maarbjerg S, Nurmikko T, Truini A, Cruccu G. Triggering trigeminal neuralgia. *Cephalalgia* 2018; 38:1049-1056.
4. Shim Y, Paeng S, Lee K, Kim S, Lee W. Trigeminal neuralgia resulting from delayed cervical cord compression after acute traumatic fracture of odontoid process. *Korean J Neurotrauma* 2019; 15:38-42.
5. Dzhafarov V, Rzaev J, Moysak G, Voronina E. Facial pain as an initial manifestation of intramedullary cervical spinal cord tumor: A case report and literature review. *Surg Neurol Int* 2020; 11:173.
6. Adachi K, Hasegawa M, Hayashi T, Nagahisa S, Hirose Y. A review of cavernous malformations with trigeminal neuralgia. *Clin Neurol Neurosurg* 2014; 125:151-154.
7. Giacobbo C, Roperto R, Cacciotti G, Mastronardi L. Cystic progression of a cavernous malformation at the level of the trigeminal root entry zone presenting with sudden onset of trigeminal neuralgia. *J Craniofac Surg* 2018; 29:e729-e730.
8. Frossand J, Domingues F, Neves P, Canhedo N, de Souza J. Cavernous malformation in the trigeminal distribution: A case report of aggressive presentation and management. *World Neurosurg* 2016; 86:514.e29-514.e22.
9. Nurmikko T, Eldridge P. Trigeminal neuralgia - pathophysiology, diagnosis and current treatment. *Br J Anaesth* 2001; 87:117-132.
10. Piovesan E, Kowacs P, Oshinsky M. Convergence of cervical and trigeminal sensory afferents. *Curr Pain Headache Rep* 2003; 7:377-383.
11. Shinozaki T, Sakamoto E, Shiiba S, et. al. Cervical plexus block helps in diagnosis of orofacial pain originating from cervical structures. *Tohoku J Exp Med* 2006; 210:41-47.
12. Woolfall P, Coulthard A. Pictorial review: Trigeminal nerve: Anatomy and pathology. *Br J Radiol* 2001; 74:458-467.
13. Ziyal I, Ozgen T. Trigeminal nerve. *J Neurosurg* 2005; 102:758-759.
14. Tubbs R, Mortazavi M, Loukas M, et. al. Anatomical study of the third occipital nerve and its potential role in occipital headache/neck pain following midline dissections of the craniocervical junction. *J Neurosurg Spine* 2011; 15:71-75.
15. Mueller O, Hagel V, Wrede K, et. al. Stimulation of the greater occipital nerve: Anatomical considerations and clinical implications. *Pain Physician* 2013; 16:E181-E189.
16. Narouze S. Cervicogenic headache. In: Benzon H, Raja S, Fishman S, et. Al (eds). *Essentials of Pain Medicine*. Elsevier, Amsterdam, 2018, pp 177-182.e1.
17. Yu M, Wang S. Anatomy, Head and Neck, Occipital Nerves. *StatPearls*. Treasure Island, Florida, 2022. www.ncbi.nlm.nih.gov/books/NBK542213/
18. Piovesan E, Kowacs P, Tatsui C, Lange M, Ribas L, Werneck L. Referred pain after painful stimulation of the greater occipital nerve in humans: Evidence of convergence of cervical afferences on trigeminal nuclei. *Cephalalgia* 2001; 21:107-109.
19. Watson D, Drummond P. Cervical referral of head pain in migraineurs: Effects on the nociceptive blink reflex. *Headache* 2014; 54:1035-1045.
20. Bendtsen L, Zakrzewska J, Abbott J, et. al. European Academy of Neurology guideline on trigeminal neuralgia. *Eur J Neurol* 2019; 26:831-849.
21. Bick S, Eskander E. Surgical treatment of trigeminal neuralgia. *Neurosurg Clin N Am* 2017; 28:429-438.
22. Yang A, Mensah-Brown K, Shektman E, et.al. Gamma Knife radiosurgery for trigeminal neuralgia provides greater pain relief at higher dose rates. *J Radiosurg SBRT* 2022; 8:117-125.
23. Zakrzewska J, Akram H. Neurosurgical interventions for the treatment of classical trigeminal neuralgia. *Cochrane Database Syst Rev* 2011; 9:CD007312.
24. Papanikolaou A, Minger E, Pais M, et. al. Management of postoperative seroma: Recommendations based on a 12-year retrospective study. *J Clin Med* 2022; 11:5062.
25. Olivo S, Magee D, Parfitt M, Major P, Thie N. The association between the cervical spine, the stomatognathic system, and craniofacial pain: A critical review. *J Orofac Pain* 2006; 20:271-287.
26. Allen R. Physical agents used in the management of chronic pain by physical therapists. *Phys Med Rehabil Clin N Am* 2006; 17:315-345.
27. Kraus S. Temporomandibular disorders, head and orofacial pain: Cervical spine considerations. *Dent Clin North Am* 2007; 51:161-193.
28. Touche R, Paris-Alemany A, Mannheimer J, et. al. Does mobilization of the upper cervical spine affect pain sensitivity and autonomic nervous system function in patients with cervico-craniofacial pain?: A randomized-controlled trial. *Clin J Pain* 2013; 29:205-215.

29. de-la-Puente-Ranea L, Gil-Martínez A, Rodríguez-Lopez O, González-Gutiérrez P, Mangas-Guijarro M, Navarro-Fernández G. Manual therapy and exercise for patients with cluster headache. *EXCLI J* 2021; 20:948-955.
30. Wechsler S. Facial pain and temporomandibular joint dysfunction secondary to acromegaly: Treatment with manual therapy, neuromuscular re-education - A case report. *Rehabil Oncol* 2020; 38:127-133.
31. Khanal D, Khatri S, Anap D. Is there any role of physiotherapy in Fothergill's Disease? *J Yoga Phys Ther* 2014; 4:162.
32. North American Spine Society Public Education Series: *Cervical Exercise: The Backbone of Spine Treatment*. 2012. pp 1-16. www.spine.org/Portals/0/Documents/KnowYourBack/CervicalExercise.pdf
33. Mirandola D, Maestrini F, Carretti G, Manetti M, Marini M. Effectiveness of an adapted physical activity protocol for upper extremity recovery and quality of life improvement in a case of seroma after breast cancer treatment. *Int J Environ Res Public Health* 2020; 17:7727.
34. Jacob T, Bracha J. Identification of signs and symptoms of axillary web syndrome and breast seroma during a course of physical therapy 7 months after lumpectomy: A case report. *Phys Ther* 2019; 99:229-239.
35. Dawson I, Stam L, Heslinga J, Kalsbeek H. Effect of shoulder immobilization on wound seroma and shoulder dysfunction following modified radical mastectomy: A randomized prospective clinical trial. *Br J Surg* 1989; 76:311-312.