Turk Neurosurg 34(3):521-523, 2024

Case Report

DOI: 10.5137/1019-5149.JTN.34594-21.1

Received: 18.03.2021
Accepted: 03.06.2021

Published Online: 16.04.2024

The Primary Afferents of Trigeminal Autonomic Reflex May not be Nociceptive: A Case Report

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ABSTRACT

Autonomic symptoms have been long noticed coming along with pain in the head, e.g. Trigeminal Neuralgia, trigeminal autonomic cephalalgias. The symptoms show up during pain attacks, so they are assumed to be activated by the nociceptive afferents of the trigeminal nerve. Here, we present a case with hypersalivation as the complication after percutaneous balloon compression for trigeminal neuralgia, although the patient was pain-free after the treatment.

A 71-year-old female with excessive salivation on the affected side after percutaneous balloon compression is described. The patient underwent microvascular decompression several years ago, and both the microvascular decompression and the preoperative imaging examination confirmed that there was no offending vessel at the root entry zone of the trigeminal nerve. After the percutaneous balloon compression, the patient was free of pain, but the autonomic symptoms (hypersalivation) still showed up. The autonomic symptoms which usually came along with pain presented solely as post-percutaneous balloon compression complication in the case.

Contrary to popular belief, for the patient who was pain-free after percutaneous balloon compression, the transiently overactivated nerve fibers that led to hypersalivation were not nociceptive afferents of the trigeminal nerve.

KEYWORDS: Trigeminal neuralgia, Autonomic symptom, Trigeminal autonomic reflex, Headache

INTRODUCTION

Excessive salivation may not be a rare symptom in trigeminal neuralgia (TN), and according to previous reports, it was occasionally seen in untreated patients during an attack of the pain. It was noted by Patrick as long ago as 1914 that vasomotor and secretory signs, like flushing, excessive salivation, nasal congestion, rhinorrhea, etc. would come along during paroxysms of trigeminal pain which could be referred to as autonomic symptoms (13,14). In previous studies, the autonomic symptoms are hypothetically attributed to pathophysiological dysfunction caused by ephaptic transmission at the root entry zone (REZ) of the trigeminal nerve (13,14), or hypothalamic activation (4). In both scenarios, the primary inputs are thought to be nociceptive afferents of the trigeminal nerve (4,13).

Percutaneous balloon compression (PTBC) is an effective treatment for TN, which mechanically compresses the gasserian (i.e. trigeminal) ganglion by balloon inflation (9). The procedure is conceived to preferentially damage the large myelinated fibers and the most common complications are numbness, dysaesthesia, paraesthesia, muscle weakness (1,3,10,11); rarely, vascular incidence (1,5). However, hypersalivation has never been reported as a complication after any surgical treatment for TN. In this report, we present a case of excessive salivation after PTBC for TN, through which we may get some insight view of the mechanism of autonomic symptoms and the wiring of associated nuclei in TN.

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CASE PRESENTATION

A 71-year-old female came to our outpatient and complained about her over secreting salvia 3 days after discharge. 4 days before, she had undergone percutaneous balloon compression (PTBC) for TN which was a recurrent shock-like facial pain on the right side consistent with the innervation territories of the II and III trigeminal nerve branches. 5 years before that, she had microvascular decompression (MVD) surgery treatment for trigeminal neuralgia which had inflicted her over 27 years. The MVD surgery only alleviated the pain, she had taken carbamazepine 600mg/d since then, and 5 years later the pain was similar to preoperative. After half a year of aggravating, she was admitted and chose the PTBC for treatment.

On admission for PTBC, the neurologic examination was normal, apart from a lightning-like pain that would go through her face when being touched the right side of her face above the corner of mouth. She has a history of hypertension and daily took Valsartan and Amlodipine Tablet, the blood pressure was 140/85 mmHg. For her pain, the patient had reached a dosage of carbamazepine 700mg/d without achieving the desired pain relief, but she suffered from severe dizziness and blurred vision. After a thoroughly preoperative examination, the PTBC was scheduled and accomplished uneventfully (Figure 1A, B). Except for postoperative numbness of the operation side, which was seen almost in every post-PTBC patient, the pain of the same side disappeared, and she was discharged 1 day later.

Three days later, as mentioned above, she presented to our outpatient with unbearable salvia over secretion. She described that she had to spit out or swallow mouthful water like salvia within every 10 seconds, and it was hard for her to fall asleep during the night that made her afraid of getting choked. The over secreting salvia came from the same side of PTBC, she recalled that, after the PTBC, it had already secreted more than usual. Vitamin B1 and B12 were subscribed for intramuscular injection. After 3 days, the symptom was much improved. At the end of 10 days follow-up, she was completely recovered from excessive salivation, and except for a little numbness of the cheek, she was free from pain.

DISCUSSION

The case is distinctive: recurrent TN, PTBC, and hypersalivation as the complication. The previous MVD surgery and preoperative MRI-TOF (Figure 1C) proved that there was no neurovascular conflict at the REZ, so that the PTBC procedure which involved trigeminal ganglion lesions could be the only cause of the hypersalivation.

In a retrospective study, Simms et al. showed that autonomic symptoms preoperatively occurred in 67 patients out of 92 TN cases (13). The autonomic symptoms include tearing, conjunctival injection, excessive salivation, sweating, facial swelling, ptosis, nasal congestion, and rhinorrhea, of which the excessive salivation is parasympathetic mediated. They proposed a pathway: nociceptive afferents are overwhelming activated at the REZ of the trigeminal nerve by ephaptic transmission, then through the connection between the trigeminal nucleus and superior salivatory nucleus, parasympathetic efferents (via the greater superficial petrosal nerve) are activated and finally lead to excessive salivation (7,13).

The REZ played a key role in Simms' theory. It has been generally accepted that the primary mechanism of classic trigeminal neuralgia is the focal demyelination of primary afferents near the REZ. The fundamental purpose of the MVD surgery is to relieve the trigeminal nerve from the offending vessels which compress on the REZ. As we have described, the case should be ruled out of vessel offending by the previous MVD and the preoperative MRI-TOF. Therefore, ephaptic transmission at the REZ was not the case here. The PTBC selectively damaged the large myelinated fibers. It is reasonable to assume that the PTBC damage is more or less like the focal demyelination at the REZ by the conflicting vessel. So, what happened at REZ could happen at the gasserian ganglion, except that the complication presented here happened during the pain was relieved, which is incompatible with the existing theory

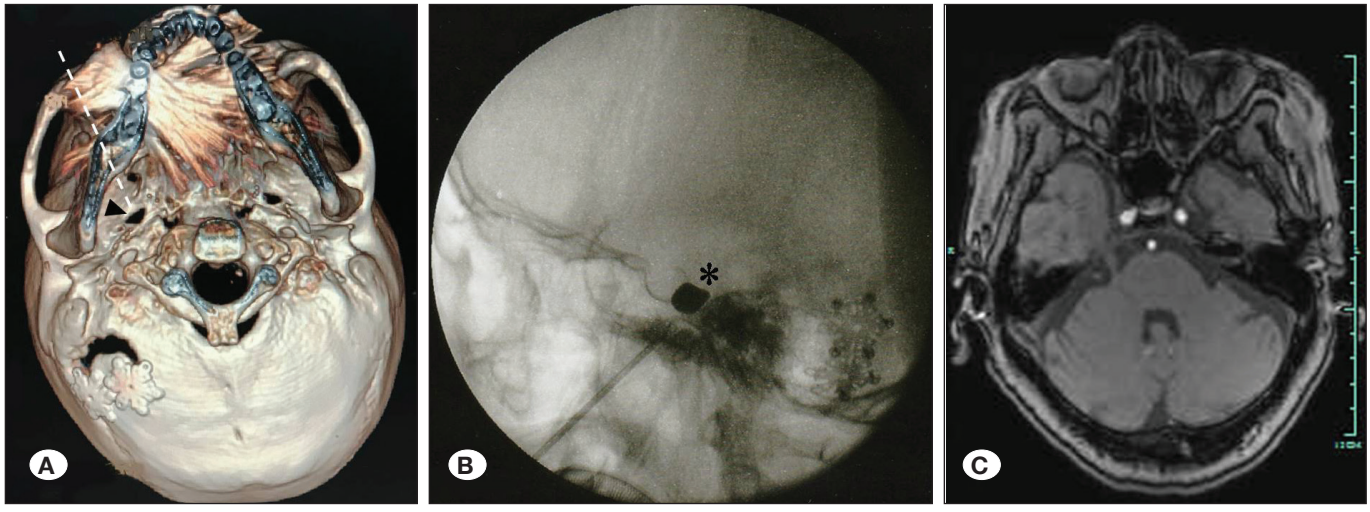

Figure 1: (A) 3D- computarized tomographt image of the trajectory and **(B)** intraoperative fluoroscopic image of PTBC, **(C)** MRI-TOF image to demonstrate there is no neurovascular involvement due to the procedure.

of pathophysiology in TN (1,8), according to which the pain should aggravate.

The autonomic symptoms are also prominent in another group of headache disorders known as trigeminal autonomic cephalalgias (TACs). The TACs includes SCUNT (short-lasting unilateral neuralgiform headache with conjunctival injection and tearing), Cluster headache, Paroxysmal hemicrania, etc (2). The pain of SCUNT typically matches the distribution of the ophthalmic branch of the trigeminal nerve and more often it is hard to differentiate the pain from TN if the TN involves the first branch (13). Trigeminal–autonomic reflex and ipsilateral hypothalamic activation are thought to play a central role in the pathophysiology of the TACs. Therapies aiming hypothalamus, such as deep brain stimulation (DBS), are proved to be helpful (4). However, hypersalivation is not a typical symptom in TACs (6,12), and tearing, rhinorrhea may be an emotional reaction in TN (Some researchers believe there are no autonomic symptoms in TN) (2). There must be a more direct connection between gasserian ganglion damage after PTBC and the hypersalivation.

We believe the case presented here was not one of a kind. During the PTBC procedure, there must be recoverable collateral damage of other nerve fibers (e.g. nerves supply touch sensory of the oral cavity) which was transiently overactivated in the process, and through the connections between the trigeminal nucleus and superior salivatory nucleus, finally led to hypersalivation. But unlike the case we presented, the damage of the related nerve fibers in PTBC usually is minimal and the subsequent salivation increase will be slight and easily to be neglected.

CONCLUSION

This is the first case in the literature demostrating hypersalivation as one of the complication of PTBC procedure. moreover, a new theory for pathway of trigeminal autonomic reflex is proposed, which is a modified version of Simms' theory: PTBC procedure transiently activates some specific nerve fibers (non-nociceptive) in the trigeminal nerve, and through the connections between the trigeminal nucleus and superior salivatory nucleus, the transient activation will lead to increasing salivation. However, the increase normally small enough to be neglected became prominent in the case we presented.

AUTHORSHIP CONTRIBUTION

Study conception and design: CS, WZ, HZ, QD
Data collection: CS, WZ, HZ, QD
Analysis and interpretation of results: CS, WZ, HZ, QD
Draft manuscript preparation: CS, WZ, HZ, QD
Critical revision of the article: CS, WZ, HZ, QD
All authors (CS, WZ, HZ, QD) reviewed the results and approved the final version of the manuscript.

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