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Case Report

A case report on probable short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing following *vairechanika nasya* in Ménière's disease

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ABSTRACT

A 62 year old woman diagnosed with Ménière's disease, who underwent *vairechanika nasya* (VN) with *shadbindu taila* presented with short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing (SUNCT) like phenomena immediately after the procedure. Rescue measures of *abhyanga* (local oil massage) and *nadi sweda* (local fomentation) were administered. Within half an hour the symptoms considerably declined and after 1 hour got completely relieved. The exact symptom disclosure by the patient who herself was a doctor helped in detecting the classic pattern of 'saw tooth phenomena' giving leads into a rare manifestation of probable SUNCT. Naranjo scale yielded zero score and thus the probable causality of VN with *shadbindu taila* could not be established so as to cause probable SUNCT as an adverse drug reaction (ADR). This case study is not put up for reporting an ADR of VN with *shadbindu taila*; rather this illustrates an uncommon, yet imperative adverse event of probable SUNCT while undergoing *nasya* procedure probably due to judgment error while fixing the VN dose in a patient with Ménière's disease. Transparent reporting of such unusual events during panchakarma procedures is necessary so that clinicians can understand, evaluate and take appropriate initiatives to manage them.

1. Introduction

Trigeminal autonomic cephalgias (TACs) are headaches characterized by trigeminal pain and autonomic signs of lacrimation and rhinorrhea with a common patho-physiology involving the trigemino-vascular system, trigemino-parasympathtic reflex and centers of circadian rhythm. TACs are classified as cluster headaches, paroxysmal hemicranias and short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing (SUNCT). SUNCT is characterized by brief paroxysmal ocular/periocular, temporal, auricular and occipital stabbing/pulsating/electric/burning attacks accompanied by ipsilateral local autonomic signs such as conjunctival injection and lacrimation. The symptoms last from 5 to 240 seconds. The attacks are yet again classified into classic single, group stabs/attacks with saw tooth pattern of attack lasting for 1 to 600 seconds i.e., upto a maximum of 10 minutes. Most commonly a SUNCT is mechanically activated wherein patients with SUNCT reveal trigeminal neuralgia like triggers. Even neck movements may trigger SUNCT [1]. Probably SUNCT is yet again a condition wherein classic clinical presentation of SUNCT is diagnosable and satisfies all but one criteria of probable SUNCT. This means, if patients have either not had a sufficient number of typical attacks of SUNCT, or have had a sufficient number but fail to fulfill one of the other criteria, it is diagnosed as a probable SUNCT [2].

Intra nasal lipid nanoparticles/liposome administration is a recently developed drug delivery system potent enough to result in an enhanced bio availability of therapeutically active molecules that demonstrate an easy access to systemic circulation with special mention to the CNS as well [3,4]. Not surprisingly, a paper highlighted significant similarities in dosage forms viz., *sneha kalpana* (therapeutic oleaginous formulations) mentioned in Ayurvedic texts and liposome drug delivery of conventional medicine in their

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origin and characterized both being lipoidal in nature [5]. Recently it has been concluded that during the intra nasal drug delivery of substances the site of deposition may influence the extent and route of absorption as well as the target organ distribution i.e. definite areas of CNS [6]. This means *nasya* is such a procedure that may possess the ability to display immediate therapeutic effects or at times abrupt untoward responses as a result of any error while administration. It is surprising that there are little published works that report any untoward event following *nasya* procedure.

This case that is being reported was admitted at National Ayurveda Research Institute for Panchakarma, Cheruthurthy (NARIP) for Ménière's disease (MD). The patient developed probable SUNCT following *vairechanika nasya* (VN), a typical category of *nasya*.

2. Case presentation

A 62 years old woman, doctor by profession who was diagnosed with MD 12 years ago came to NARIP for better care. She was on vertin 16 mg for vertigo like symptoms since diagnosed with MD. Medical history revealed that she was consuming medications for dyslipidemia as well. Based on symptoms as per Ayurvedic sciences, MD in this patient was co-relatable to *sannipatha sopha* in the inner ear (edematous occurrence). She was presented with a chronic history of symptoms of vertigo associated with transient hearing loss (with episodes of vertigo), tinnitus and fullness of ears. The symptoms were significantly co-relatable to *bhrama* (*vataja nanatmaja/vata-pittaja*), *karnanaada* (*vataja*), *badhirya* (*vataja/vata-kaphaja*) and *karnapratinaha* (*vataja/vata-kaphaja*). Thus *vata kaphottara sannipata* (*Vata*+++, *Kapha*++, *Pitta*+) involvement in manifestation of the disease (*sopha*) was inferred.

Treatment procedures including *dipana*, *pachana* (metabolism correcting internal medicaments), *snehapana* (intake of medicated ghee intended to facilitate therapeutic purgation) and *virechana* (therapeutic purgation) were administered in the patient and subsequently VN was planned. VN refers to a specific modality of *nasya* wherein relatively potent formulation is being intra nasally administered so as to have an enhanced bio-availability of medicaments bringing about definite purificatory efficacies. The most common form of medication utilized for this purpose is *taila* (medicated oils) under *sneha kalpana*. In this case *shadbindu taila*, a poly herbal formulation was used [7].

VN was planned at 10 o'clock in the morning. Pre operative procedures of VN such as *abhyanga* (oil massage) and *nadi sweda* (medicated fomentation) were performed. The dose of *shadbindu taila* was fixed as six ml (three ml for each nostril). As per SOP of the Institute the dose of 3 ml per nostril was planned to be instilled in a divided dose of 1.5 ml (i.e. in two bouts). The untoward event occurred at the end of the second bout of nasal instillation (remaining 1.5 ml) in the right nostril.

Patient developed severe burning excruciating pain in the right frontal region, right occipital region (verbal rating scale-VRS score 9) along with redness and lacrimation from the ipsilateral eye. There was no neck stiffness, nausea, vomiting and photophobia. Her vitals were stable. Patient was advised to remain on the treatment table for 15 minutes. But the symptoms (VRS 9) persisted. As reported by the patient, there were around 30 attacks each lasting for 20 seconds during this eventful 1 hour and there was lack of complete relief in pain between attacks-indicating a saw-tooth phenomenon for the event. *Abhyanga* and mild *nadi sweda* over nape of neck, occiput and forehead as rescue measures were reiterated. Within half an hour the symptoms considerably declined (VRS 3) and after one hour got completely relieved (VRS zero). No further episodes were reported on the same day. The chronological course of the event is summarized in Table 1.

3. Discussion

When pathological changes in MD were considered, it was understood that following anoxia of stria vascularis capillaries (which secrete endolymph), there was significant increase in capillary permeability which result in enhanced transudation of fluid and thereby increase endolymph production leading to distension of endolymphatic sac.

Considering the symptoms of *badhirya* [Deafness], *karnanaada* [ringing in the ear] and *karna pratinaha* [fullness in the ears], the site of disease manifestation was considered to be *sthanika* (localized) i.e., in the inner ear. MD is fundamentally an endolymphatic hydrop disorder. Thereby the condition was significantly co-relatable to *udakavaha srotodushti* [channels of body fluids] ([12], *Shareera Sthana*; *Siravibhaga*: Chapter 6 verse 24) at the level of *shabdavaha sira* [inner ear channels conducting sound] ([12] *Shareera Sthana*; *Siravibhaga*: Chapter 6 verse 8). The pathogenesis is best co-relatable to *sira aayama* [distension of channels] (*sopha poorva rupa* [prodromal features of edema]) ([12] *Nidana Sthana*; *panduroga kamala sopha visarpa nidanam*: Chapter 13 verse 16) of *udakavaha srotodooshita shabdavaha sira* followed by *sira tanutva* [Increased permeability of channels] [9, Chikitsa Sthana, Swayathu chikitsitham, Chapter 12 verse 11], *utsedha* [Elevation] and *samhata* [Compact] ([12] *Nidana Sthana*; *panduroga kamala sopha visarpa nidanam*: Chapter 13 verse 11) which are the classic symptoms of *sopha* [edema]. The symptoms manifested here were co-relatable to *vata kaphottara sannipata*. VN is indicated in sopha ([12] *Sutra Sthana*; *Nasyavidhi*: Chapter 29 verse 5). Thus VN with *shadbindu taila* in *marsha matra* [relatively higher doses] was selected here as the treatment of choice.

Untoward event of probable SUNCT was observed following VN administration in the patient's right nostril. Based on the Naranjo Adverse Drug Reaction Probability Scale [14], the score at that instance was zero which made *shadbindu taila* a doubtful cause for an adverse drug reaction (ADR) of probable SUNCT. Thereafter probable SUNCT was considered as an adverse event (AE) while VN in the patient.

Complications in the form of headache may manifest in VN due to various reasons involving faulty administration such as errors in dosage, increased temperature of the medicament administered etc ([12] *Sutra Sthana*; *Nasyavidhi*: Chapter 29 verse 5)]. We ruled out all these probabilities with a critical analysis of time to time documentation of events as a part of SOP of documentation and detailed history taking. But the fact remains that there is an increased possibility of a judgmental error that might have occurred with respect to initial VN dosage with *shadbindu taila* in this case of Ménière's disease in particular. At the first instance, when the event occurred, it was thought that there was an involvement of disrupted *vata* and *pitta doshas* in the event. An opinion on doing *ksheera nasya* (*pittahara* management) as a rescue modality surfaced whichcould not be materialized due to unavailability of required ingredients. Further, the patient complained about severe pain and discomfort. Thus pain management through *abhyanga* and a little amount of *nadi sweda* over nape of neck, occiput and forehead were administered.

Differential diagnosis between occipital neuralgia, trigeminal neuralgia and all varieties of TACs were scrutinized and it is depicted in Table 2. Analyzing the degree of autonomic stimulation as evident by the degree of pain (VAS-9) along with the presence of 'the saw tooth phenomenon' associated with ipsilateral lacrimation and redness of eyes gave leads into diagnosing the phenomena of probable SUNCT.

Now, all possible triggers for probable SUNCT namely mechanical stimulation (neck posture while *nasya*, *abhyanga*), trigeminal triggers (faulty suction, high dose of potent medication) and

Table 1
Timeline of the case reported.

2009	Patient had an episode of severe vertigo and vomiting. Symptomatic management with conservative medicine.
2011	Similar complaints recurred, consulted an ENT surgeon. Diagnosed with mild deafness in left ear and associated vertigo. Symptomatic management for vertigo. There was an erratic pattern of recurrence and remission till 2017.
2017	Frequent episodes of vertigo, vomiting with ringing sound in the left ear. She was diagnosed with Ménière's disease and initiated treatment for the same. However, her symptoms worsened incapacitating her to perform day to day activities. She underwent ayurvedic treatment from elsewhere.
15-04-2021	Admitted in our hospital for further management. Oral medicines started. *Punarnavadi kashaya* ([8], *Kashaya kalpana*) (15 ml added to 45 ml luke warm water twice daily before food), *Chandraprabhavati* [9] and *Suvarnamuktadi gulika* (One tablet each twice daily along with decoction)
22-04-2021 to 24-04-2021 (3 days)	*Udwarthana* (whole body massage with medicated powder) with *Kolakulathadi churna* ([10], *Sutra Sthana*, *Aragwadheeyam adhyayam*, Chapter 13 verse 118)
25-04-2021 to 27-04-2021 (3 days)	*Abhyanga* with *Kshirabala taila* ([11], *Chikitsa Sthana*, *Vatashonita chikitsitham*, Chapter 22 Versa 45–46) and *Nadi sweda* *Vicharana snehapana* with *Kshirashadpala ghrira* ([11], *Chikitsa Sthana*, *Rajayakshmadi chikitsitham*, Chapter 5 Versa 22–23) (30 g with rice gruel in lunch time)
28-04-2021	*Virechana* with *Gandharvahastadi eranda taila* ([12], *Chikitsa Sthana*, *Vidradhivrdhichikitsa*, Chapter 15, Verse 25.) - 30 ml at 6.00 AM
29-04-2021 to 05-05-2021 (7days)	*Vairechanika nasya* with *shadbindu taila*- 3 ml each nostril in the morning. But due to the probable SUNCT episode on day one, reduced to 2 ml each nostril for subsequent days. *Thalam* (applying medicated paste over bregma) with *kshirabala taila* and *Rasnadi churna* ([8], *Churna Prakarana*) *Karna dhoopana* with *Laksha* (slick serum from *Laccifer lacca)*, *Guggulu* (*Commiphora wightii* (Arn.) Bhandari), *Kushta* (*Saussurea lappa* (Decne.) Sch.Bip.) and *Karpura* (*Cinnamomum camphora* (L.) J.Presl.) [13] in the evening.
06-05-2021 to 12-05-2021 (7 days)	*Takradhara* (pouring of medicated butter milk over scalp in a rhythmic fashion) ([8], *Dhara kalpa*) in the morning *Karnapurana* (medicated fumigation into ear canal) with *Devadaru taila* ([13], *Uttara Sthana*, *Karnaroga Pratishedham* chapter 18 verse 5) in the evening. Episode of vertigo on 08-05-2021 in the morning. Got relieved in 15 minutes.
September 2021	On telephonic follow up, the patient complained of frequent episode of vertigo even after full course of treatment from this institute. Took ENT consultation and was on Tab Vertin 48 mg and Tab Diamox twice daily. Since she had no relief, cardiac and neurological evaluation was conducted. Was under Tab Gabapentin 200 mg twice daily. But still experinced sudden vertigo attack in between. Her MRI findings on 18-09-2021 showed bilateral anterior inferior cerebellar artery (AICA) loop–AICA loop within the left internal acoustic canal extending >50% into the IAC (internal acoustic canal) (Type III Chavda). AICA loop in the right cerebello-pontine angle bordering the right internal acoustic meatus and not entering the IAC (Type I Chavda).She is under further investigations to find out the cause for vertigo, in a higher medical center.

olfactory afferent irritation were considered. It was intriguing that the patient didn't develop similar episodes in the following six days of VN. However, it should be noted that the treatment was modified (3 ml per nostril was reduced to 2 ml per nostril in the subsequent days and the patient was once again educated regarding do's and don'ts while *nasya* administration) for the remaining days of VN.

Another curious fact found while follow up was regarding the MRI brain report of the patient which was taken following her discharge from NARIP. Her report showed bilateral AICA (Anterior inferior cerebellar artery) loop. AICA loop in the left entered the internal acoustic canal while that of the right bordered the right internal acoustic meatus and did not it. Varied sorts of vascular compressions of the trigeminal nerve could manifest as probable SUNCT [15]. Though in this case the AICA loop causing trigeminal compression precipitating in SUNCT is not causally ascertainable but it is plausibly co-relatable. Further radiological investigations may ascertain this phenomena. Thus the diagnosis of probable SUNCT is even more likely here.

As per Ayurveda, considering the mode of action of *nasya*, *sringataka marma* is the target area mentioned for the medicaments ([12], *Sutra Sthana*; *Nasyavidhi*: Chapter 29 verse 3). *Sringata marma* considering it to be a *dhamani marma* (simulating area of blood flow) within the cranial cavity, it is best translated as cavernous sinus [16]. Cavernous sinus has proximity with the internal carotid artery. It is found that drugs absorbed from the mucosa beyond the nasal valve drains via veins that end up in cavernous sinus where the blood may come in contact with the carotid artery [6]. To elaborate further, it may be noted that perivascular neurogenic inflammatory process of the internal carotid artery in its bony canal can trigger TACs and cluster headaches [1].

VN in terms of its potency and administered dose can plausibly result in enhanced mucosal absorption, antidromic stimulation of nociceptive fibers [17] and subsequent irritation of olfactory and trigeminal afferents. This can eventually result in initiation of certain perivascular neurogenic inflammatory processes triggering TAC. Here in this case, the existing AICA loop which is relatively capable of triggering vascular compression over trigeminal nerve may have played a catalyst role that eventually lead to the abrupt incidence of the phenomenon.

This report highlights the fact that fixing correct dose of nasya medicament for VN is crucial to avoid such untoward events. In this particular case of Ménière's disease, as cited above; we felt a possible judgmental error that might have occurred while fixing the initial dose of VN. Hence we suggest that a trial run may be carried out with lower dose levels/*Pratimarsha nasya* followed by carefully analyzing dose response by systematically increasing the dose especially in patients with a history of cephalgias of any origin or in symptoms of orthostatic hypotension/vertigo/tinnitus/Ménière's disease etc should be included in the treatement SOP.

Table 2
Differential diagnosis of the case with respect to symptoms presented.

	Cluster head ache	Paroxysmal hemicrania	SUNA	SUNCT
Burning excruciating pain	–	–	+	+
Cutaneous trigger	–	–	+	+
Autonomic features	+	+	+ (but redness and lacrimation is absent)	+

SUNCT – short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing.
SUNA – short-lasting unilateral neuralgiform headache attacks with cranial autonomic symptoms.

4. Conclusion

This case study reports an uncommon AE of probable SUNCT during administration of VN in a patient of Ménière's disease. The importance of detailed history taking connecting various symptoms and presentations, astute documentation of every event occured during *nasya* procedure or as a matter of fact in any *panchakarma* procedure, expecting specific untoward events along with analysis of its dosha status and keeping ready rescue medications based on dosha status are very crucial. Moreover, reporting of such unusual events is necessary so that so that clinicians can understand, evaluate and take appropriate initiatives to manage them.

Author contributions

V. Krishna Kumar-Writing - Managed the case, Prepared Original Draft. Pratibha. P. Nair- Writing - Review & Editing, Methodology. G.N. Sree Deepthi-Conceptualization. Pradeep Kumar P. P-Conceptualization.

Conflict of interest

None.

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Appendix A. Supplementary data

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