Snoring-Induced Vibratory Angioedema

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Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Patient: Female, 70
Final Diagnosis: Snoring induced vibratory angioedema
Symptoms: Swelling of tongue • roof of mouth and throat • multiple episodes at night
Medication: —
Clinical Procedure: Continuous positive airway pressure therapy
Specialty: Allergology

Objective: Rare disease
Background: Vibratory angioedema (VA) is a rare physical urticaria, with symptoms of itching and swelling of the skin or mucosa when it is exposed to vibration. Avoidance of vibration is the best way to manage this condition. This case report will assist physicians to diagnose this rare condition. Here, a previously unpublished potential successful treatment modality is being presented, with good symptom control, along with some photographs taken during an acute attack. A literature review points towards potential undiagnosed cases.
Case Report: A 70-year-old woman had multiple emergency department visits for tongue and throat swelling over 3 years. The episodes always happened at night. Detailed history elicited some episodes of itching and swelling of hands when driving as well as significant snoring while sleeping. Physical examination was unremarkable except for morbid obesity. Complement factor 4 and C1esterase inhibitor level were within normal limits. A tentative diagnosis of angioedema induced by oropharyngeal vibration from snoring was made. A sleep study confirmed sleep apnea with severe snoring. After CPAP (continuous positive airway pressure) treatment, she had successful symptom control.
Conclusions: Snoring-induced VA is very likely an under-diagnosed condition in the community. The typical history is the key to the diagnosis. This condition could be confirmed by vibration test or by the resolution of symptoms with elimination of vibration. Effective symptom control is possible by avoidance of oropharyngeal vibration from snoring with the administration of CPAP therapy, making it a potential novel indication for this condition.

MeSH Keywords: Angioedema • Sleep Apnea, Obstructive • Snoring • Urticaria

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Background

Vibratory angioedema (VA) is a rare physical urticaria, with symptoms of itching and swelling of the skin or mucosa when it is exposed to vibration [1,2]. Some common triggers are mowing the lawn and riding a motorcycle; at-risk occupations include jack-hammer operator, [3] carpenter, machinist, and metal grinder [4]. Mast cell degranulation is the proposed pathophysiological mechanism, but is not always observed [3]. Autosomal-dominant inheritance pattern [5] and non-familial cases have been described [5,6]. Avoidance of vibration is the best way to manage this condition [3,4]. This case report will assist physicians to diagnose this rare disorder when it is encountered. A previously unpublished potential successful treatment modality is presented, with good symptom control, along with some photographs taken during an acute attack. A literature review indicates many potential undiagnosed cases.

Case Report

A 70-year-old woman had multiple emergency department visits for tongue and throat swelling over 3 years. These episodes always happened at night, which would wake her from sleep and caused difficulty in swallowing. There were no other associated symptoms. The symptoms progressed slowly over several hours and then improved during the next 2 days. These episodes happened approximately every 3 months but became progressively more frequent, culminating in 2 attacks during the week before her first visit. Detailed history elicited some episodes of itching and swelling of hands when driving, as well

as significant snoring while sleeping. Her active medical diagnoses included allergic rhinitis, chronic obstructive pulmonary disease, coronary artery disease, hypertension, hypothyroidism, and gastro-esophageal reflux disease. She lived in Michigan, wintered in Arizona, and had these episodes in both places. Allergy testing showed sensitivity to cats and molds but she had no cats at home. No family members had similar symptoms. Of note, she had gained 60 lbs. during the previous 10 years. Her complete medication list was Albuterol inhaler, Clonazepam, Clopidogrel, Fluticasone Propionate/Salmeterol inhalation diskus, Gabapentin, Isosorbide Mononitrate, Levothyroxine, Paroxetine, and Propranolol. Physical examination was unremarkable except for morbid obesity. C4 (complement factor 4) was 37 mg/dl (14–40 mg/dl) and C1 esterase inhibitor level was 28 mg/dl (19–37 mg/dl); these were measured during an acute attack. A CT scan of soft tissue of the neck, which was taken during one of the attacks, showed diffuse prominence of the right parapharyngeal soft tissue structures, which measured about 1.5 cm in anteroposterior and transverse diameter, extending for approximately 5 cm from the level of the uvula to a level equal to the upper margin of the arytenoid cartilage region and displaced the uvula to the left side. The process was well defined without evidence of central necrosis or any drainable fluid collection. The hyoid bone and the epiglottis were not involved. There were no adjacent inflammatory changes. Tongue and soft palate images were significantly limited because of artifacts from dental fillings.

Since these episodes always happened while sleeping, in addition to presence of snoring history and hand swellings while driving (vibration of the steering wheel), a tentative diagnosis

Figure 1. Localized swelling on the left lateral tongue.

Figure 2. Lateral view of the tongue swelling.

of angioedema induced by vibration of the soft palate, tongue, and pharyngeal wall from snoring was made. A sleep study showed 2 apneas and 26 hypopneas in a 5 hour 22 minute sleep period, producing an apnea-hypopnea index of 5.2 (<4). Her Snore index was 46.4%, with oxygen desaturation down to 85%. After CPAP (continuous positive airway pressure) treatment with 10-cm pressure support, she only had 1 episode of VA during the next 6 months, which occurred when her mask was out of position. Her tongue swelling during this episode was mild and is illustrated in Figures 1 and 2. This was followed by 1 more attack when she was noncompliant with CPAP for a week.

Discussion

The diagnostic test recommended for VA by a European consensus panel is by reproduced edema on the forearm by induced vibration using a laboratory vortex mixer run at 780–1380 rpm (rotations per minute) for 10 minutes [7].

The C4 and C1 esterase inhibitor level tests were not suggestive of hereditary angioedemas. Vortex mixer testing, as mentioned above, was not performed because of the typical history elicited and the lack of a vortex mixer in my clinic.

The aim of CPAP therapy here is to decrease or prevent snoring, thereby eliminating vibration of tongue, soft palate, and pharyngeal wall, thus preventing VA. Complete symptom control was achieved in this patient with CPAP therapy other than

during the instances when the mask was displaced accidentally and when the patient was noncompliant with CPAP. This confirmed the diagnosis and efficacy of the proposed management.

On review of the literature, there was a prospective study conducted over 2 years on edema of the uvula. The sample size was 58 patients, out of which 32 (55%) had idiopathic etiology. Comparative statistical analysis showed that 28 (87%) out of the 32 idiopathic cases had snoring (P value <0.0001) [8]. These idiopathic cases could be undiagnosed snoring-induced VAs.

Conclusions

Snoring-induced vibratory angioedema is very likely an under-diagnosed condition in the community. The typical history is the key to the diagnosis. This condition could be confirmed by vibration test or by the resolution of symptoms with elimination of vibration. Effective symptom control was possible in this patient by avoidance of oropharyngeal vibration from snoring with the administration of CPAP therapy. Not all patients who snore have sleep apnea; hence, oropharyngeal vibratory angioedema from snoring could be an indication by itself for CPAP therapy.

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