Patient-Reported Factors Associated with the Onset of Hyperfunctional Voice Disorders

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Abstract

Objectives: Prevention, diagnosis, and treatment of hyperfunctional voice disorders would be improved by better understanding their etiological contributing factors. Therefore, this study estimated the prevalence of etiological factors using self-reported data about disorder onset from a large cohort of patients with Phonotraumatic Vocal Hyperfunction (PVH) and Non-Phonotraumatic Vocal Hyperfunction (NPVH).

Methods: Retrospective chart review extracted the self-reported rate (gradual, sudden) and events associated (voice use, anxiety/stress, upper respiratory infection [URI]) with disorder onset from 1,577 patients with PVH and 979 patients with NPVH.

Results: Both patient groups reported a gradual onset more than a sudden onset. Voice use was the most frequently reported event for PVH and the NPVH group self-reported all three events at equal frequency. The largest PVH subgroups were associated with voice use while the NPVH subgroups were associated with only voice use, only URI, or only anxiety/stress.

Conclusion: The results support the general clinical view that PVH is most strongly related to the gradual accumulated effects of phonotrauma, while NPVH has a more heterogeneous etiology. The identified PVH and NPVH subgroups may have clinical relevance and future work could investigate differences in treatment and outcomes among these subgroups.

Keywords

table fold nodules, muscle tension dysphonia, voice disorders, vocal hyperfunction

Introduction

Voice disorders have been estimated to affect approximately 6.6% to 7.6% of adults in the United States at any given point in time1,2 and the most commonly treated voice disorders are associated with vocal hyperfunction.3 Vocal hyperfunction (VH) refers to chronic conditions of abuse and/or misuse of the vocal mechanism due to excessive and/or unbalanced muscular force.3 Generally, VH can be divided into two subtypes: Phonotraumatic Vocal Hyperfunction (PVH) and Non-Phonotraumatic Vocal Hyperfunction (NPVH).4 PVH is associated with obvious signs of true vocal fold trauma (eg, bilateral vocal fold nodules, polyps). Non-Phonotraumatic Vocal Hyperfunction—also referred to as functional dysphonia5 or primary muscle tension dysphonia6—is associated with symptoms such as chronic dysphonia and vocal fatigue in the absence of vocal fold tissue trauma. Because NPVH is a diagnosis of exclusion (ie, no physical or neurological disorder can account for the vocal symptoms), this disorder is associated with a myriad of potential etiologies (eg, stress, anxiety, upper respiratory infections/URI, voice overuse).6-15 All voice disorders and putative contributing factors are standardly labeled according to the Classification Manual for Voice Disorders.16

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The chronic VH state is thought to be caused by factors such as transient periods of increased vocal demands with inadequate vocal recovery time (eg, prolonged loud talking), increased psychological stress promoting non-volitional (autonomic nervous system) heightened activity of the perilymphageal and/or extralaryngeal muscles; and environmental factors like URLs, laryngopharyngeal reflux (LPR), irritants, and dehydration. Additionally, there are some commonly held clinical notions that various eliciting factors are more likely to be associated with PVH or NPVH. For example, increased voice use is believed to play a larger role in PVH than in NPVH and increased stress and anxiety are assumed to play a more prominent role in NPVH than PVH.

Current knowledge regarding the causative factors of hyperfunctional voice disorders relies mostly on clinical impressions and empirical data acquired after disorder occurrence (measures of vocal function, personality/psychological profiles, etc). Causative theories, when informed by data collected after disorder formation, contain a high degree of uncertainty because one cannot separate whether the measurements of interest pre-existed the voice disorder and/or characterize compensatory behaviors due to the disorder. To objectively evaluate causative factors, the gold standard would be a prospective observational study of subjects in a high-risk occupation.

This study design is pragmatically impossible for behaviorally-based voice disorders as the incidence is very low, even in high-risk occupations (estimates 5-7%). However, as a starting point, patients can be asked to recall what they thought was associated with the onset of their voice disorder or how long they think it took for their disorder to occur. Since, to the author’s knowledge, no study has formally analyzed self-report data regarding what the patient believed was associated with their disorder onset, this study attempted to gain a better understanding of the relative prevalence of etiologic factors for PVH and NPVH and to use this information to identify potentially useful clinically meaningful subgroups. We hypothesized that (1) patients from both PVH and NPVH will report gradual onsets more often than sudden onsets (supporting the hypothesized habitual nature of the disorders); (2) patients with PVH would self-report voice use as the most frequent event associated with voice disorder onset and all large subgroups will be associated with voice use (supporting “phonotrauma” as the putative causative factor of PVH); and (3) the NPVH group would equally self-report all associated events and all subgroups will be unidimensional (supporting the hypothesis that this group is etiologically heterogeneous).

Methods

Participant Recruitment

All research was approved by the hospitals’ Institutional Review Boards. Patients with a primary diagnosis of vocal fold nodules, vocal fold polyp(s), or muscle tension dysphonia were retrospectively identified in an automated search of the clinical database from the Massachusetts General Hospital Center for Laryngeal Surgery and Voice Rehabilitation (MGH Voice Center) from the years 2006 to 2017 and a deidentified copy of the database from the Massachusetts Eye and Ear Infirmary (MEEI) Voice and Speech Laboratory from the years 1994 to 2004. Diagnoses were based on a comprehensive team evaluation (laryngologist and speech-language pathologist) at the MEEI or MGH Voice Center that included (1) the collection of a complete case history, (2) endoscopic imaging of the larynx and (3) instrumental aerodynamic and acoustic assessment of vocal function. After performing the automatic database search, each patient was included or excluded after manually reading their laryngology evaluation. Patients with NPVH (ie, primary muscle tension dysphonia) were included in the study if they had secondary diagnoses of LPR and gastro-esophageal reflux disease (GERD). However, patients with NPVH were excluded if they had any secondary diagnoses related to structural or neurological disorders: specifically laryngitis, loss of superficial lamina propria, benign lesion, polyp, cyst, dysphagia, sulci, paradoxical vocal fold motion, any mention (confirmed or possible) of upper airway paralysis or paresis, polypoid corditis, keratosis, presbylarynx, fibrovascular changes, leukoplakia, injury of the recurrent or superior laryngeal nerve, or history of radiation or neurological impairment.

Patients with PVH (ie, vocal fold nodules and polyps) were included in the study if they had secondary diagnoses of LPR and GERD as well as those commonly associated with
phonotrauma, specifically: erythema, edema, varices, ec-
tasia, laryngitis, secondary/reactive muscle tension dysphonia, hemorrhage. However, patients with PVH were excluded if they displayed secondary diagnoses not related to phono-
trauma, specifically: cyst, pseudocyst, unilateral (ie, the lesions had to be bilateral), sulci, cancer, bamboo nodule, anterior web, or paradoxical vocal fold motion.

**Case History Form**

The case history form was filled out after the laryngology evaluation and before the speech-language pathology evaluation. It included two questions related to factors associated with the onset of the patient’s voice disorder:

1. Was the onset of your voice disorder: sudden, gradual, unsure, or not applicable (circle the most appropriate response).
2. Were there any events or circumstances that occurred with the onset of your voice difficulty: Anxiety/stress, increased voice use, vocal abuse (yelling/screaming), URI, change in job, injury (trauma), surgery, chemical exposure, accident, none, other, and not applicable (circle all that apply).

All responses to the case history form questions were coded as binary variables: 1 = the patient selected the response; 0 = the patient did not select the response.

**Statistics and Analysis**

Differences between the PVH and NPVH groups were evaluated for four onset variables (ie, gradual, sudden, unsure, not applicable), six associated event variables (ie, voice use, anxiety/stress, URI, other, not applicable, and none), six combinations of variables to investigate interactions between onset*event (ie, gradual or sudden onset and voice use, anxiety/stress, or URI), and (3) six patient subgroups (anxiety/stress only, voice use only, URI only, anxiety/stress and voice use, voice use and URI, and other only). Only three individual associated events were statistically compared because Voice Use and Vocal Abuse were combined into one category labeled “Voice Use” and the categories of Voice Use, Anxiety/Stress, and URI were the only ones to be reported by at least 10% of patients. All other associated events were grouped into the “Other” category. Two (group) by two (presence/absence of category) Chi Square tests were performed to evaluate pair-wise between-group differences (PVH vs NPVH). Significance was Bonferroni-corrected to the level of $P < .002$ for each of the 22 total $2 \times 2$ comparisons because of multiple hypothesis testing. Odd’s ratios (OR) were used as an effect size metric to help represent the potential clinical significance of any statistically significant pair-wise between-group differences. Patient subgroups were identified when approximately 100 or more patients reported only a single event or the same combination of multiple events.

**Table 1. Number of Patients (Percentage of Total Patients) that Reported Duration of Onset Occurrence Within Each Group.**

<table>
<thead>
<tr>
<th>Onset Time Course</th>
<th>Patient Groups</th>
<th>Odds Ratios</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>PVH</td>
<td>NPVH</td>
</tr>
<tr>
<td>Gradual</td>
<td>902 (57%)</td>
<td>422 (43%)</td>
</tr>
<tr>
<td>Sudden</td>
<td>321 (20%)</td>
<td>293 (30%)</td>
</tr>
<tr>
<td>Unsure</td>
<td>218 (14%)</td>
<td>135 (14%)</td>
</tr>
<tr>
<td>Not applicable</td>
<td>136 (9%)</td>
<td>129 (13%)</td>
</tr>
</tbody>
</table>

**Table 2. Frequency of Patient-Reported Associated Events with the Onset of Disorder Within Each Group.**

<table>
<thead>
<tr>
<th>Patient Reported Associated Event</th>
<th>Patient Groups</th>
<th>Odds Ratios</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>PVH</td>
<td>NPVH</td>
</tr>
<tr>
<td>Anxiety/Stress</td>
<td>291 (19%)</td>
<td>181 (19%)</td>
</tr>
<tr>
<td>Gradual onset</td>
<td>176 (11%)</td>
<td>100 (5%)</td>
</tr>
<tr>
<td>Sudden onset</td>
<td>60 (4%)</td>
<td>48 (5%)</td>
</tr>
<tr>
<td>Voice use</td>
<td>850 (54%)</td>
<td>227 (23%)</td>
</tr>
<tr>
<td>Gradual onset</td>
<td>544 (35%)</td>
<td>116 (12%)</td>
</tr>
<tr>
<td>Sudden onset</td>
<td>165 (11%)</td>
<td>57 (6%)</td>
</tr>
<tr>
<td>URI</td>
<td>307 (20%)</td>
<td>212 (22%)</td>
</tr>
<tr>
<td>Gradual onset</td>
<td>175 (11%)</td>
<td>93 (10%)</td>
</tr>
<tr>
<td>Sudden onset</td>
<td>83 (5%)</td>
<td>82 (4%)</td>
</tr>
<tr>
<td>Other</td>
<td>290 (18%)</td>
<td>316 (32%)</td>
</tr>
<tr>
<td>None</td>
<td>108 (7%)</td>
<td>124 (13%)</td>
</tr>
<tr>
<td>Not applicable</td>
<td>466 (20%)</td>
<td>317 (15%)</td>
</tr>
</tbody>
</table>

**Results**

The automatic database search identified 3,105 total patients with a diagnosis of PVH or NPVH. After manual filtering, 2,556 were included in the analysis: 1,577 patients with PVH and 979 patients with NPVH.

Table 1 shows the rate of onset for all groups. While both groups reported gradual onset more often than sudden onset, gradual onset was reported more frequently by patients with PVH (57%) than NPVH (43%); $\chi^2(1)=48.0, P < .001$, OR = 1.8. Sudden onset was reported more frequently by patients with NPVH (30%) than those with PVH (20%); $\chi^2(1)=30.3, P < .001$, OR = 1.7.

Table 2 shows the events associated with disorder onset. The most frequently reported event for both groups was...
The purpose of this study was twofold: (1) to gain a better understanding of the relative prevalence of etiologic factors for PVH and NPVH and (2) to identify clinically meaningful patient subgroups. The patient-reported information supported multiple commonly-held clinical notions about the similarities and differences in disorder onset between the two types of hyperfunctional voice disorders. For example, it is thought that PVH and NPVH are associated with chronic, long-term vocal behaviors and the patients in both groups more frequently self-reported their disorder onset as gradual (~50%) than sudden (~25%). Furthermore, when patients estimated in their case history form how long the symptoms of their disorder were present before seeing a laryngologist, most reported months (49%) or years (46%) and very few reported a timescale of weeks or less (5%).

Patient self-report data supported the clinical belief that PVH is mainly caused and/or associated with habitual, chronic voice use—specifically the accumulated effect of phonotrauma. Over half of the PVH group (54%) reported voice use was associated with the beginning of their symptoms and 32% of the PVH group reported that voice use was the only event associated with disorder onset. Additionally, three out of four patient subgroups included voice use as an associated event. The PVH subgroups have potential to guide research into more refined conceptual models or frameworks of how phonotraumatic lesions are formed. For example, although the biggest patient subgroup reported only voice use and nothing else associated with disorder onset, the next two largest subgroups (voice use and anxiety/stress; voice use and URI) suggest that excessive phonotrauma may not only result from too much voice use. A large portion of phonotraumatic lesions may form during periods of vocal fold tissue degradation (URI) in the presence of “typical levels” of phonotrauma. This is further reinforced by the smallest subgroup where the only associated event was URI. Also, as supported by the Voice Use and Anxiety/Stress subgroup, perhaps increased phonotrauma can occur because of anxiety- or stress-provoking situations (eg, shouting during arguments or social events). In the patient subgroups where Voice Use is not the sole contributor, prevention and treatment approaches focused on changing how the patient generally produces voice would probably be less effective than vocal hygiene education or practicing “healthy/efficient” voicing under emotional situations.

Results for patients with diagnoses of nodules and polyps were combined in the PVH category. This was done for two reasons. First, diagnoses of patients in the databases were determined by several different clinical teams that could have employed different criteria for differentiating between nodules and polyps, which are both associated with phonotrauma and can sometimes be challenging to definitively classify (eg, differentiating bilateral nodules vs a polyp and reactive nodule). Second, there were no differences between the groups of patients diagnosed with nodules or polyps that were meaningful enough to change the final interpretation of the results, particularly with respect to comparisons with the NPVH group.

As was expected, the self-report data from the NPVH group were heterogeneous. The NPVH group equally reported Voice Use, Anxiety/Stress, and URI (~20%) as associated with the onset of their disorder; and the largest

### Table 3. Frequency of Combinations of Patient-Reported Associated Events with the Onset of Disorder Within Each Group.

<table>
<thead>
<tr>
<th>Associated Onset Patient Clusters</th>
<th>Patient Group</th>
<th>Odds Ratios</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>PVH</td>
<td>NPVH</td>
</tr>
<tr>
<td>Anxiety/Stress only</td>
<td>50 (3.2%)</td>
<td>98 (10.0%)</td>
</tr>
<tr>
<td>Voice use only</td>
<td>505 (32.0%)</td>
<td>124 (12.7%)</td>
</tr>
<tr>
<td>URI only</td>
<td>107 (6.8%)</td>
<td>130 (13.3%)</td>
</tr>
<tr>
<td>Anxiety/Stress and URI</td>
<td>21 (1.3%)</td>
<td>25 (2.6%)</td>
</tr>
<tr>
<td>Anxiety/Stress and voice use</td>
<td>166 (10.5%)</td>
<td>46 (4.7%)</td>
</tr>
<tr>
<td>Voice use and URI</td>
<td>125 (7.9%)</td>
<td>45 (4.6%)</td>
</tr>
<tr>
<td>Anxiety/Stress, voice use, and URI</td>
<td>54 (3.4%)</td>
<td>12 (1.2%)</td>
</tr>
<tr>
<td>Other only</td>
<td>124 (7.9%)</td>
<td>226 (23.1%)</td>
</tr>
</tbody>
</table>

Odds ratios with * or “a” represent a higher prevalence in PVH or NPVH, respectively.

Notes: “n/a” is denoted for patient groupings that did not have a large enough sample size for reliable Chi Square analysis. Abbreviations: NPVH, nonphonotraumatic vocal hyperfunction; PVH, phonotraumatic vocal hyperfunction; URI, upper respiratory infection. P < .001, OR = 3.9, respectively. There was no significant difference in self-reported anxiety/stress or URI between the NPVH and PVH groups. The group-based differences (or lack thereof) remained unchanged after separately evaluating gradual and sudden onsets associated with voice use, anxiety/stress, or URI.

Table 3 shows all unique patient-reported combinations of associated events. Besides the large amount of patients who reported no associated events, not applicable, or only “Other,” there were four large PVH subgroups (Voice use only = 505, Voice use and anxiety/stress = 166, Voice use and URI = 125, and URI only = 107), two large NPVH subgroups (URI only = 130 and Voice use only = 124), and one borderline large NPVH subgroup (Anxiety/stress only = 98). Three of the four PVH subgroups occurred more frequently in the PVH group than the NPVH group: Voice use only, $\chi^2(1) = 121.9$, $P < .001$, OR = 3.2; Anxiety/stress and voice use, $\chi^2(1) = 30.0$, $P < .001$, OR = 2.4; Voice use and URI, $\chi^2(1) = 10.8$, $P < .001$, OR = 1.8. Two of the three NPVH subgroups occurred more frequently in the NPVH group than the PVH group: URI only, $\chi^2(1) = 30.3$, $P < .001$, OR = 2.1; Anxiety/stress only, $\chi^2(1) = 51.8$, $P < .001$, OR = 3.4.
subgroup reported only an “Other” event. The “Other” events consisted of many infrequently (<100 patients across both PVH and NPVH groups) reported events like various surgeries, dysphagia, reflux, allergies, and job change. Furthermore, this heterogeneity was mostly distributed in a unidimensional way, as 65% of patients with NPVH reported only one category of associated onsets. The large NPVH subgroups were also unidimensional, as 10% of patients only reported Anxiety/Stress, 12% only reported Voice Use, and 13% only reported URI. Perhaps these findings indicate that, in general, there are three subtypes of NPVH patients; all of which have been mentioned sporadically throughout the existent literature. The “Voice Use” subgroup (representing the traditional view of primary muscle tension dysphonia”) could be described as those who use their voice a great deal, but are not vocally efficient enough to fulfill their daily needs without vocal fatigue and/or deterioration. The “URI” subgroup could be described as those who had no vocal difficulties until their upper respiratory system was physically compromised. The vocal compensations used to communicate during the URI became habitual and remained despite recovery from the infection. Recent literature suggests that upper respiratory symptoms were associated with a 49% to 75% increased odds of developing a voice disorder in general. Finally, the “Anxiety/Stress” subgroup could be described as those whose vocal function deteriorates in association with a psychological state or event that impacts voice production. There are multiple commonly-used diagnostic labels and theories/frameworks in the literature specifically referring to a psychological association with NPVH (eg, psychogenic, functional). These three subgroups could also indicate the need for different behavioral treatment approaches; for example, improving the efficiency of voicing for the “Voice Use” subgroup, helping patients access the voice they had before their infection for “URI” subgroup, and addressing how psychologically stressful or anxious events relate to their voice disorder in the “Anxiety/Stress” subgroup.

The chronic nature of these disorders and their relationship to events in daily life reinforce the clinical importance of developing assessments that consider longer time periods than traditional in-clinic measurements of vocal function/behavior. Ambulatory voice monitoring technology can provide estimates of how patients are using their voice in daily life over extended time scales (eg, hours, days, weeks). Additionally, the events or situations associated with disorder onset can be difficult to replicate during a therapy session; for example, artificially evoking a realistic level of stress is hard. Therefore, ambulatory voice monitoring could potentially measure vocal behavior in real-life stressful situations for assessment purposes or to provide biofeedback to assist with maintaining good vocal function during stressful events.

A limitation of the study is that the data consist entirely of patient self-report. However, this is the closest empirical measurement possible without directly observing subjects before they experience any vocal difficulty. It is also possible that the “Voice Use” event is over-reported since the patients’ disorders inherently affected their voice. All patients had seen a laryngologist and were given a diagnosis before the case history form was filled out, which could have influenced their responses. However, the large number of subjects in each group should mitigate the effect of noisy self-report metrics.

**Conclusion**

A large cohort of patients with PVH (n=1,577) and NPVH (n=979) provided self-report data on the onset of their voice disorder in terms of rate (sudden vs gradual) and associated events. The patient self-report data support the general clinical view that PVH is most strongly related to the gradual accumulated effects of phonotrauma, while NPVH has a more heterogeneous etiology. Subgroups of PVH and NPVH patients were identified that may have clinical relevance and future work could investigate differences in treatment and outcomes among these subgroups.

**Declaration of Conflicting Interests**

The author(s) declared the following potential conflicts of interest with respect to the research, authorship, and/or publication of this article: Drs Robert Hillman and Steven Zeitels have a financial interest in InnoVoyce LLC, a company focused on developing and commercializing technologies for the prevention, diagnosis, and treatment of voice-related disorders. Dr Hillman’s and Dr Zeitels’ interests were reviewed and are managed by Massachusetts General Hospital and Partners HealthCare in accordance with their conflict of interest policies.

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