Development and Validation of the Voice Catastrophization Index

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Summary: Introduction. Catastrophization is a cognitive distortion that has been studied in pain patients and found to be a significant factor in their disability and response to treatment. Dysphonia patients may demonstrate a similar behavior, suggesting the existence of voice catastrophization.

Objective. To establish the validity of the Voice Catastrophization Index (VCI), a new instrument estimating voice symptoms catastrophization.

Methods. A prospective study. Patients with and without voice conditions were administered with the VCI, Voice-Related Quality of Life (V-RQOL), and the Generalized Anxiety Disorder-7 (GAD-7)questionnaires. Consensus Auditory-Perceptual Evaluation of Voice (CAPE-V) rating was determined for patients with voice conditions.

Results. Ninety-one patients participated, 65 with voice conditions and 26 without. 61.5% were females; mean age was 49.8 ± 15.7 years. The VCI score for patients with voice conditions was significantly higher, 22.46 ± 16.56, compared with 3.96 ± 10.79, respectively (P value < 0.0001). The VCI demonstrated moderate correlation with both V-RQOL and GAD-7 scores: −0.562 and 0.560, respectively (P value < 0.000001); however, it showed no correlation with the CAPE-V. The VCI's internal consistency with each of its three dimensions and 13 items was acceptable to strong. The reproducibility and stability was demonstrated in a subgroup of 26 patients; 81% of these patients had a difference of 10 or less points between the two evaluations.

Conclusions. The VCI accomplished the requirements of a scale's validity for estimation of voice symptoms catastrophization in voice patients. The potential role of voice catastrophization as a predictor of treatment response and tailoring can now be investigated using the VCI.

Key Words: Dysphonia–Catastrophization–Voice Catastrophization Index–Anxiety–Quality of life.

INTRODUCTION

Catastrophization has been described for more than a half century as a phenomenon of cognitive distortion that might, yet not necessarily, be associated with anxiety and depressive disorders.[1] Catastrophization in individuals who experience pain was originally studied[2] in 1979 and was defined as concern, fear, and inability to divert attention from pain. In the following years, other properties of pain related catastrophization were investigated, including tendency to exaggerate or magnify the painful situation,[3] as well as helplessness and pessimism regarding the ability to cope with the pain.[4] Subsequently, in 1995, the Pain Catastrophizing Scale (PCS) was developed by Sullivan et al.[5] The PCS contains 13 items subgrouped under three dimensions: rumination, magnification, and helplessness. With rumination, patients are consumed by their thoughts regarding their symptoms. With magnification, patients exaggerate the impact of their symptoms, often ascribing to themselves a much poorer prognosis. Helplessness is when patients feel like nothing could ever alter their situation. Since then, the PCS was validated in multiple languages[6–10] and for various pain conditions.[11–15] As a result, the phenomenon of catastrophization in pain patients was well investigated and translated to clinical implications. In pain patients, catastrophization was found to be associated with physical disability[1] and to correlate with poorer prognosis and decreased effectiveness of therapy.[16] Recently, the clinical use of catastrophization scales was extended to other somatosensory conditions in addition to pain.[17] In particular, the role of catastrophization is currently investigated in patients with vestibular disorders.[17–20]

Although associated with other psychological conditions, catastrophization has been shown to be a distinct phenomenon from anxiety and depressive disorder. Studies on coping strategies and chronic pain adjustment found that catastrophizing was related to poorer coping and adjustment, even when controlling for depression level was applied.[21]

Patients experiencing pain catastrophization are known to have poorer quality of life (QOL) along with high rates of disability.[22,23] A study on 1208 patients with chronic pain[23] investigated the effect of different variables on the patients' QOL using a multiple regression analysis; the analysis demonstrated pain catastrophizing to be the single most important predictor of QOL in these patients. Specifically, social functioning, vitality, mental health, and general health were significantly associated with pain catastrophizing. Nevertheless, catastrophization is distinct from QOL, because it describes the patient's inner world, thoughts, and beliefs, whereas the concept of QOL follows the patient's outer world and general well-being with respect to different aspects of life and interaction with the environment.

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It can be assumed that the phenomenon of catastrophization can be found also in voice patients. Patients with voice conditions were found to have higher prevalence of anxiety, depression, personality, and other psychiatric disorders.[24–26] Moreover, the cognitive state was found to influence voice patients' response to treatment. A research on benign causes for dysphonia and anxiety has demonstrated that high Clinical Anxiety Scale scores correlated with incomplete vocal recovery with treatment.[27] These studies, however, did not distinguish catastrophization from anxiety and depression. Voice symptoms, similarly to pain symptoms, can be magnified by perception abnormalities, with continuous thoughts about the voice problem, and helplessness about finding resolution. In our clinical experience, we have observed voice patients in whom the physical findings and QOL instruments poorly correlate with the load of their voice symptoms.

Although there is some evidence suggesting voice catastrophization exists, no studies have been performed to evaluate its impact, partially because of the lack of a validated instrument. Given pain catastrophization's independence from QOL, it is reasonable that voice catastrophization is independent from the currently used scales for measuring QOL or handicap related to voice condition. Designing this study, we assumed that the missing piece for clarifying the discrepancy between the true voice quality and the patients' perception of their voice is the degree in which these patients catastrophize their voice symptoms.

Therefore, in this study, we aimed to produce and validate a new tool to estimate the degree of catastrophization in patients with voice conditions. We also sought to examine the relations of this new tool with anxiety and the existing voice-related QOL scale. Validating this new tool can establish the foundations for future investigation on the role of catastrophization in voice patients and their response to treatment.

METHODS

Scale production

The 13 items of the PSC were adapted and modified to apply to voice complaints. These adapted items formed together the Voice Catastrophization Index (VCI) as presented in *Box 1*. Similarly to the PCS, the VCI is a 13-item scale with statements representing the three dimensions of catastrophization; items 1, 2, 3, 4, 5, and 12 represent helplessness; items 8, 9, 10, and 11 represent rumination; and items 6, 7, and 13 represent magnification. For each item, the patient is asked to indicate the degree to which he or she experiences these thoughts and feelings on a 0–4 scale. The maximal possible score is 52, and the minimal score is 0; a higher score represents higher catastrophizing of the voice condition.

Study design

A prospective validation pilot study was conducted from May 2015 to August 2015, at the Emory Voice Center at Emory University Hospital Midtown, a large tertiary care medical center. After securing institutional review board approval, 91 patients, from all patients who visited the laryngology clinic, were recruited to participate in the study.

The inclusion criterion for the case group (patients with voice condition) was all new adult patients with either clinically perceived dysphonia or a patient-reported voice complaint. The voice conditions in this group include but not limited to the following: neurologic disorders such as spasmodic dysphonia, vocal cord paralysis and paresis, phonotraumatic lesions, primary muscle tension dysphonia (MTD), laryngitis, and laryngeal malignancy. The control group (patients without voice conditions) included all new adult patients presenting to the Voice Center without voice complaints (eg, complaints of dysphagia or cough), without dysphonia perceived by the clinicians, and no evidence of voice

Box 1
The Voice Catastrophization Index. (VCI) Listed below are statements describing thoughts and feelings that may be associated with voice conditions. Please indicate the degree to which you have these thoughts and feelings using the scale below. 0 = Not at all; 1 = To a slight degree; 2 = To a moderate degree; 3 = To a great degree; 4 = All the time

#		0	1	2	3	4
1	I continuously worry whether my voice problem will ever go away.					
2	I feel I can't go on with my voice like it is.					
3	My voice is terrible and I think it's never going to get any better.					
4	My voice problem is awful and I feel that it overwhelms me.					
5	I feel I can't stand my voice problem anymore.					
6	I become afraid that my voice problem will get worse.					
7	I keep thinking of other times I have had voice problems.					
8	I anxiously want the voice problem to go away.					
9	I can't keep my voice problem out of my mind.					
10	I keep thinking about how much trouble my voice problem gives me.					
11	I keep thinking about how badly I want my voice problem to stop.					
12	I feel that there is nothing I can do to reduce the intensity of my voice problem.					
13	I wonder whether something serious may happen.					

condition during examination. Patients were excluded from the study if they refused to participate or if they were unable to understand or complete the required study questionnaires.

Patients who agreed to participate in the study were properly consented and administered with the following three questionnaires: the VCI, the Voice-Related Quality of Life (V-RQOL),[28] and the Generalized Anxiety Disorder-7 (GAD-7).[29,30] Patients read and answered the surveys themselves without direct intervention or assistance, unless it was requested by the patient. For patients with voice conditions (the case group), a standardized voice recording was performed during that visit. The voice recording protocol included reading of a standardized passage, sustained phonation of the vowels /a/ and /i/, load phonation of the word "Hey," and a glissando from low to high note. These voice recordings were used to determine the patient's Consensus Auditory-Perceptual Evaluation of Voice (CAPE-V)[31] rating. A team of two speech and language pathologists exclusively treating voice disorders, blinded to the patient's identity, characteristics, and study group, rated each voice sample according to the CAPE-V. Furthermore, 20% of the samples were scrambled and randomly repeated to ensure intrarater reliability. Overall, for each participant, data collection included age, gender, chief complaint, diagnosis, comorbidities, psychiatric history, the VCI, V-RQOL, and GAD-7 scores, and for the case group also the CAPE-V score.

To examine test-retest reproducibility, a subgroup of the study patients were administered the same three scales for the second time within a 2-week period of the initial visit. Patients were excluded from this subgroup analysis if they had a subjective voice change, per patient or clinician, or had any intervention taken place between the two tests (eg, patients with acute laryngitis in whom treatment was initiated immediately after the initial visit were excluded). For those patients who did not have a visit scheduled within 2 weeks, the same scales were administered by a study investigator via online survey. Patients who had no visit scheduled within 2 weeks and were unwilling or unable to provide online survey responses were also excluded from this subgroup analysis.

Statistical analysis

Based on standards put forth by the Medical Outcomes Trust in 1995, validation of a scale must satisfy requirements of validity, reliability, responsiveness, and low burden.[32] To show validity, face validity and construct validity were examined. To examine face validity, the VCI was evaluated by three fellowship-trained laryngologists, who are also familiar with the PSC and the concept of catastrophization. For construct validity, VCI's ability to discriminate between patients with and without voice conditions was tested via case-control comparison using Mann-Whitney *U* test. Furthermore, the VCI was analyzed for correlation or independence from the GAD-7, V-RQOL, and CAPE-V scores. To test correlation between each two fixed interval scoring systems, Pearson correlation coefficient was used. To show reliability, internal consistency and test-retest reproducibility were examined. For determination of internal consistency, Cronbach alpha correlation coefficients were calculated for each of the three dimensions and the 13 items of the VCI. A correlation coefficient of at least 0.6 was considered acceptable and at least 0.7 was considered good. Finally, the test-retest reproducibility was analyzed by comparing global VCI scores at the initial visit to the VCI scores on the second visit using Mann-Whitney *U* test. Finally, to show appropriately low burden, the time and load needed to administer and complete the questionnaire were considered. For all statistic tests performed, an alpha of 5% or less was considered to be statistically significant. Statistical analyses were accomplished using *SPSS Statistics* 20.0 (IBM, Armonk, NY).

RESULTS

A total number of 91 patients were recruited for the study: 65 with voice conditions (case group) and 26 patients without voice condition (control group). Fifty-six of the participants (61.5%) were females, and the mean age was 49.8 ± 15.7 (range, 18–80 years). Twelve percent (n = 11) of the patients reported pain associated with their chief complaint and 23% (n = 21) had prior diagnosis of either anxiety or depression. Selected characteristics of the study population by group are shown in Table 1. The VCI was self-administered by all patients; none of the patients had to ask for assistance and none reported any difficulty with the survey.

Validity

Face validity was satisfied by assessment of the VCI by three fellowship-trained laryngologists who are also familiar with the PSC and the concept of catastrophization. The laryngologists agreed that the VCI appears to measure voice catastrophization. With respect to construct validity, the total VCI score for patients with any voice condition was significantly higher from the score for patients without voice condition, 22.46 ± 16.56, compared with 3.96 ± 10.79, respectively (*P* value < 0.0001) (Table 2). Correlations between the VCI scores and the V-RQOL, CAPE-V, and GAD-7 scores according to Pearson correlation coefficient are presented in Table 3. The VCI demonstrated moderate significant correlation with both V-RQOL and GAD-7 scores: –0.562 and 0.560, respectively (*P* value < 0.000001). The VCI showed no correlation with the CAPE-V (0.088; *P* value = 0.491).

Reliability and internal consistency

The internal consistency of the VCI based on the scale's three dimensions and 13 items is presented in detail in Table 4. The total VCI score strongly correlated with both the helplessness (items 1, 2, 3, 4, 5, and 12) and the magnification (items 6, 7, and 13) dimensions, with Cronbach alpha correlation coefficients of 0.934 and 0.792, respectively. For the rumination dimension (items 8, 9, 10, and 11), the Cronbach alpha was acceptable at 0.620.

Reliability and reproducibility

A subgroup of 26 study patients participated in further investigation of test-retest reproducibility. Of the 65 patients with voice conditions, three patients were withdrawn as they transferred their care to other facilities; another 36 patients were excluded from this investigation as they fulfilled at least one of the exclusion

TABLE 1.
Select Demographics, Medical History, Complaints, and Condition of the Study Population by Group

	Patients With Voice Condition (Case Group)	Patients Without Voice Condition (Control Group)	*P* Value
Total number	65	26	
Age mean ± SD (range)	52.4 ± 14.9 (20–77)	43.5 ± 16.1 (18–80)	0.02*
Female, N (%)	40 (61.5%)	16 (61.5%)	1.00†
Pain associated with the complaint N (%)	8 (12.3%)	3 (11.5%)	0.920†
Anxiety or depression history N (%)	14 (21.5%)	7 (26.9%)	0.582†
Diagnosed disorder at the initial visit N (%)	Primary muscle tension dysphonia 19 (29%) Vocal fold paralysis or paresis 13 (20%) Phonotraumatic lesion 8 (12%) Atrophy 7 (11%) Dystonia or tremor 6 (9%) Laryngitis 6 (9%) Other 6 (9%)	Cough 6 (23%) Dysphagia 6 (23%) Recurrent tonsillitis 2 (8%) Obstructive sleep apnea 2 (8%) Other 10 (38%)	

* *P* value calculated by unpaired Student *t* test.
† *P* value calculated by square chi-square test.

TABLE 2.
Scoring of VCI, VRQOL, and GAD-7 by Group

		Patients With Voice Condition (Case Group)	Patients Without Voice Condition (Control Group)	*P* Value
VCI	Mean ± SD	22.46 ± 16.51	3.96 ± 10.79	<0.00001 *
	Median (IQR)	18 (7–39)	0 (0–2)	(U, 167)
V-RQOL	Mean ± SD	61.88 ± 28.03	97.12 ± 6.55	<0.00001 *
	Median (IQR)	65 (40–87.5)	100 (97.5–100)	(U, 89)
GAD-7	Mean ± SD	7.4 ± 6.16	5.62 ± 4.86	0.3125 *
	Median (IQR)	6 (2–12)	5 (2–9.5)	(U, 729.5)

* *P* value calculated by Mann-Whitney *U* test.

TABLE 3.
Correlation Between the VCI Scores and the V-RQOL, CAPE-V, and GAD-7 for Patients With Voice Condition

		V-RQOL (N = 65)	CAPE-V (N = 64)	GAD-7 (N = 65)
VCI total	Pearson correlation coefficient	–0.562*	0.088	0.560*
	Sig. (two-tailed)	0.000001	0.491	0.000001

* Correlation is significant at the 0.05 level (two-tailed).

criteria described in the Methods section. Eventually, 26 patients with voice condition who had no subjective voice change, and no immediate intervention, were reevaluated for the second time within a 2-week period. The test-retest reproducibility was analyzed by comparing the VCI total score at the initial visit with the VCI score on the second visit. The patients' V-RQOL scores remained relatively stable between those two visits: a change from 60.0 ± 30.7 to 61.4 ± 23.5 (*P* value = 0.850). The total VCI

TABLE 4.
Analysis of the Internal Consistency of the VCI by Dimensions and Individual Items

Dimension	Dimension to Total VCI Correlation (Cronbach Alpha)	Item	Item to Total VCI Correlation (Cronbach Alpha)
Helplessness	0.934	1	0.719
		2	0.853
		3	0.858
		4	0.923
		5	0.904
		12	0.577
Rumination	0.620	8	0.643
		9	0.412
		10	0.673
		11	0.685
Magnification	0.792	6	0.709
		7	0.512
		13	0.698

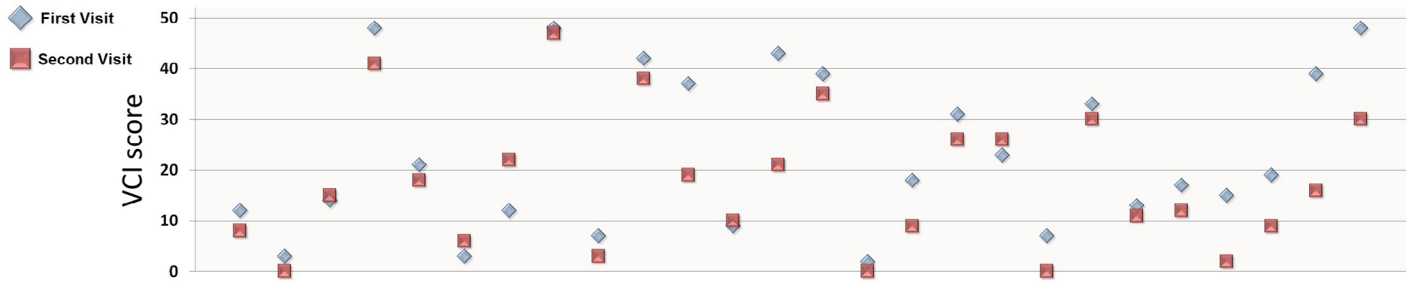

FIGURE 1. Reproducibility of the VCI in 26 patients with no treatment and no subjective voice change (each individual patient is represented on a different location on the horizontal axis).

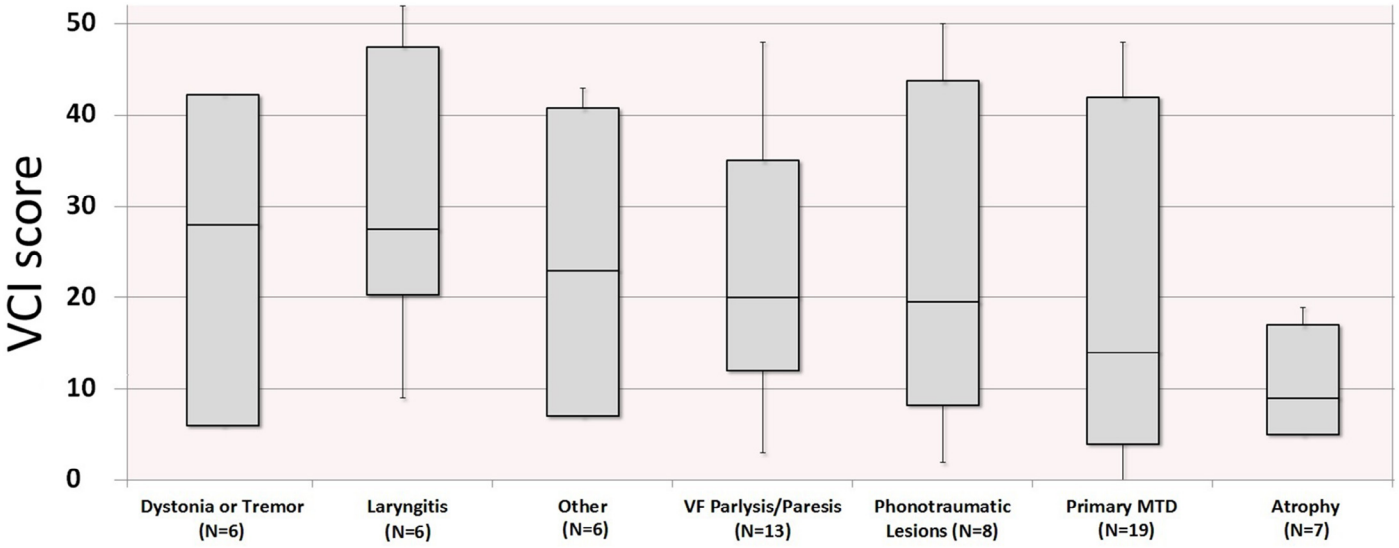

FIGURE 2. Box plots describing the VCI score by voice disorder groups. Whiskers and boxes for: range, interquartile range, and median.

score also demonstrated a relative stability, with no statistically significant difference; a mean total score of 23.2 ± 15.5 on the first visit compared with 17.5 ± 13.4 on the second ($P = 0.161$). Eighty-one percent (n = 21) of those patients had a difference of 10 or less points between the two evaluations. The reproducibility of the VCI, as demonstrated in each of those 26 patients with no treatment and no subjective voice change, is presented in Figure 1.

Catastrophization by patients' characteristics

In the group of 65 patients with voice conditions, a univariate analysis showed that the VCI score was not significantly affected by any of the following variables: gender (22.5 ± 17.1 *versus* 22.4 ± 16.1, P value = 0.968), age over 60 years (18.6 ± 16.3 *versus* 24.9 ± 16.5, P value = 0.131), symptoms duration over 1 year (22.7 ± 17.0 *versus* 21.4 ± 15.1, P value = 0.841), associated pain (26.9 ± 17.9 *versus* 21.8 ± 16.4, P value = 0.430), and previous diagnosis of depression or anxiety (24.4 ± 17.8 *versus* 21.9 ± 16.4, P value = 0.697). The VCI scores by disorder groups are presented in Figure 2.

DISCUSSION

Hereby we originally present the concept of catastrophization in voice patients along with a new instrument to measure it, the VCI. In this study, the VCI accomplished the various requirements of a scale's validity, reliability, and responsiveness. VCI scores were able to distinguish patients with voice conditions from those without (by significantly higher scores). Furthermore, as would be expected from a catastrophization scale, the VCI scores failed to correlate with a relatively reliable measurement of voice quality (CAPE-V), whereas it moderately correlated with both anxiety (GAD-7) and QOL instrument (V-RQOL). The VCI showed high internal consistency (acceptable to strong correlations) with each of its 13 items and each of the three dimensions of catastrophization (helplessness, magnification, and rumination). Moreover, when the same patients were asked to answer the VCI on different occasions, the scores were comparable, indicating the scale's reproducibility. Overall, these properties of the VCI make it a reliable instrument that can be used for further investigation of the role of catastrophization in voice patients.

Originally, when developing the VCI, the study assumed it could be used to confirm diagnosis or identify functional voice disorder or primary MTD, because functional dysphonia was associated with anxiety and depression on previous studies.[33,34] However, it was found that catastrophization is relatively common among patients with organic voice problems, and the VCI may serve for other clinical applications. Our preliminary data

(Figure 2) failed to support the assumption that patients with functional voice disorder or primary MTD have higher tendency for catastrophization. This might be related to the small sample size, and should be further investigated before drawing conclusions. Similarly, the univariate analysis in our study failed to demonstrate significant association between voice-related catastrophization and various patients' characteristics: gender, older age, duration of symptoms, associated pain, or diagnosis of depression or anxiety. Yet the aim of our current study was to validate the VCI, and therefore its sample size is insufficient for proving or excluding these possible associations; further investigation of a larger cohort with multivariant analysis is required. Although insignificant, a suspected trend of reduced voice catastrophization among elderly patients was noticed (P value = 0.131). In correspondence, patients with atrophy tended to have lower voice catastrophization compared with patients with other voice conditions (Figure 2). These findings can be attributed either to age-related catastrophization trend or to the typical slow progression of vocal fold atrophy. Nonetheless, these are merely nonsignificant preliminary findings, which should encourage further investigation.

In our study, the VCI showed moderate correlation with the GAD-7 scale; this supports our hypothesis that the VCI is an instrument that reliably estimates catastrophization of voice conditions. This correlation implies that high VCI scores are associated with anxiety, which is a well-known feature of somatosensory catastrophization.[35] Yet because this is only a moderate correlation, it can be assumed that VCI measures an entity different from anxiety. Anxiety, depression, and other psychiatric conditions were found to have higher prevalence among patients with voice disorders[24–26] and to correlate with treatment outcome.[27] However, these studies did not estimate the catastrophization component in those patients; therefore, the possibility of confounding by catastrophization cannot be excluded. The use of the VCI will allow investigation of these states and the impact of catastrophization on treatment outcomes.

The VCI scores in our current cohort moderately correlated with the V-RQOL; this corresponds with what was already established in catastrophizing pain patients.[23] Nevertheless, the fact that this was merely a moderate correlation strengthens our notion that voice catastrophization, as represented by the VCI, is a distinct entity from QOL. Although the commonly used scales for voice-related QOL and handicap (V-RQOL and Voice Handicap Index) focus on vocal everyday tasks (talking over the phone or in a noisy room, "trouble doing my job" etc), and interaction with others (going out socially, "feel annoyed when people ask me to repeat" etc), the VCI focuses on cognition and the internal aspects of disability. The items presented by the VCI focus on cognitive distortion of the voice condition, which eventually translates into disability. We believe that voice catastrophization by the means of VCI can provide finer resolution of voice-related disability, which sometimes can massively occupy the patient's internal world with only minor changes on the outside. Moreover, functional and social aspects of voice problems, as described by QOL and handicap scales, may alter between cultures and different personalities; nevertheless, the items included in the VCI describe cognitive states, and as such can be considered more universal, and less liable by situational changes. Nonetheless, the authors do not argue that the VCI can replace voice-related QOL and handicap scales for measurement of treatment response, as cognition is unlikely to be affected by treatment addressed for voice conditions.

There are several possible clinical applications to voice catastrophization that, now with the availability of a validated instrument, can be examined. In general, a mismatch between the physical impairment and what one would expect from the physical findings should raise a suspicion for a catastrophizing state, and the VCI can be used to confirm or refute the suspicion. More specifically, when making a decision on treatment for patients with organic voice condition, the clinician anticipates the pretreatment voice-related QOL or handicap scales to demonstrate disability that will improve with appropriate treatment. Yet some patients may not demonstrate improvement in these scales following treatment, whereas more reliable measurements for voice quality (eg, CAPE-V) improve.[36] We argue that these are patients experiencing catastrophization and can be recognized as such before initiation of treatment, using the new validated tool, the VCI. Using voice-related QOL or handicap scales alone would fail to identify these patients, as their voice-related QOL before treatment is likely to be poor regardless of their degree of catastrophization. Hence, the VCI can be valuable in identifying patients with catastrophization, which are less probable to improve with conventional treatment. Acknowledging the catastrophization state would allow the physician to address the patient's needs in a more global manner and discuss reasonable expectations. To achieve response to treatment, it might be appropriate to encourage a patient experiencing catastrophization to participate in a support program or cognitive therapy. Moreover, it is possible that alterations in VCI scores can be used independently to guide the timing for initiating treatment. These assumptions should be verified in future studies.

Even though this publication provides key insights regarding the role of catastrophization in the management and outcome of voice patients, we are still far from completely understanding it. Our current study is limited by its relatively small sample size aimed to establish validation alone; results presented in our current study regarding patient's and disease characteristics by VCI should be considered preliminary. Future studies using the VCI will be able to verify or exclude our assumption that catastrophization plays a significant role in the disability and response to treatment of voice patients. Furthermore, future studies will be able to investigate other clinical applications of the VCI, such as identifying patients with functional dysphonia. These future investigations can now be accomplished after the VCI instrument was validated.

CONCLUSIONS

The VCI is a validated tool for estimation of the degree of voice-related catastrophization in voice patients. The suggested clinical applications of the VCI correspond with the well-established clinical applications in pain medicine. The potential role of voice catastrophization as a predictor of response to treatment and treatment tailoring for the individual patient can now be investigated using the VCI.

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