

## Two-dimensional Scheme for Arranging Voice Disorders

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**IMPORTANCE** Prevalent schemes that have been used for arranging voice pathologies have shaped theoretical and clinical views and the conceptualization of the pathologies and of the field as a whole. However, these available schemes contain inconsistencies and categorical overlaps.

**OBJECTIVE** To develop and evaluate a new approach for arranging voice pathologies, using 2 continuous scales, organicity and tonicity, which were used to construct a 2-dimensional plane.

**DESIGN, SETTING, AND PARTICIPANTS** This survey study was conducted among experts in the fields of laryngology and/or voice disorders from 10 countries. The survey was conducted using an online platform from March to May 2021. The data were analyzed in June 2021. Of the 45 experts who were initially approached, 39 (86.7%) completed the survey.

**MAIN OUTCOMES AND MEASURES** The primary outcome measures were group ratings on 2 rating scales: organicity and tonicity. On the organicity scale, 0 represented nonorganic and 10 organic. On the tonicity scale, 0 represented hypotonic and 10 hypertonic.

**RESULTS** Participants included 16 laryngologists and 23 speech-language pathologists, of whom 27 (69.2%) were women and 12 (30.8%) men with a mean age of 55 years. The Cronbach  $\alpha$  was high for organicity and tonicity (0.98 and 0.97, respectively). Interrater agreement ( $r_{wg}$ ) was moderate to very strong ( $r_{wg} \geq 0.50$ ) for most pathologies. The correlation between the 2 scales was moderate and negative ( $r = -0.38$ ;  $P = .03$ ). The pathologies were scattered across the full range of both scales and the 4 quadrants of the 2-dimensional plane, suggesting the continuity and bidimensionality of the new arrangement scheme. In addition, a latent profile analysis suggested that the 4-cluster solution is valid and roughly corresponded to the 4 quadrants of the constructed plane.

**CONCLUSIONS AND RELEVANCE** The findings of this survey study suggest the potential use of a 2-dimensional plane that was based on 2 continuous scales as a new arrangement scheme for voice disorders. The results suggest that this approach provides a valid representation of the field based on 2 basic measures beyond the specific etiology of each laryngeal pathology or condition. This simple and comprehensive organization scheme has the potential to facilitate new insights on the nature of voice pathologies, considering the interpathology similarities and differences.

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Generally, theoretical schemes for arranging clinical pathologies within any medical field mold the conceptualization of researchers and clinicians of the different pathologies and the discipline as a whole.<sup>1</sup> Therefore, the importance of arranging pathologies and, in the current context, voice pathologies into a simple and comprehensive scheme is crucial for theoretical thinking and clinical practice. Various organization schemes for voice pathologies have been proposed. The most basic divides voice disorders into 2 major taxonomic categories, based on their etiology: organic vs functional.<sup>2</sup> Within this scheme, neurological voice disorders are included in the organic category, yet others regard them separately from structural pathologies.<sup>3</sup> The second major category, functional, includes both pathologies that stem from excessive muscle tension and pathologies with various psychogenic origins.<sup>1,2,4</sup> An alternative binary scheme has also been proposed that separates voice disorders based on laryngeal muscular activity. This scheme defines 2 categories of voice pathologies: *hypofunctional* and *hyperfunctional*.<sup>5</sup>

Because many clinicians have viewed these binary schemes as limited or rigid, more elaborate categorization systems have been proposed. For example, a 4-category scheme has been suggested in which voice disorders are divided according to their etiology into (1) congenital pathologies, (2) pathologies of the vocal fold cover, (3) neurogenic pathologies, and (4) pathologies of muscular dysfunction.<sup>6</sup> Others proposed more detailed schemes, for example, defining 7 categories: (1) structural, (2) inflammatory, (3) trauma or injury, (4) systemic, (5) digestive, (6) neurological, and (7) psychological.<sup>7-9</sup>

To our knowledge, all available categorization schemes postulate that a specific voice pathology/condition fits into a unique category. Hence, the possibility of an overlap between the different categories, or the realization that some pathologies may fit into more than a single category, is viewed as a theoretical pitfall rather than an inherent feature.<sup>2,4,7,8</sup> An example of the limitations of these categorical schemes is the fact that the diagnoses of psychogenic aphonia and muscle tension dysphonia are categorized as *functional*, although their origins and their clinical manifestation could be very different. Another common example of the overlap between these categories is *vocal nodules*, which are typically described as resulting from a combination of organic and behavioral (ie, functional) factors.<sup>2-6</sup>

In light of these limitations of the available categorization schemes, and because of the complex nature of many voice disorders, this study was designed to examine whether an alternative approach can be developed for arranging vocal dysfunctions that would better represent the natural distribution of voice disorders. The proposed approach is based on the realization that even the very basic categories are not mutually exclusive.<sup>9</sup> Thus, we sought to examine the possibility of using 2 intersecting continuous scales: organicity and tonicity. To this end, *organicity* is defined as a continuum that ranges between organic and nonorganic, and *tonicity* is defined as a continuum that ranges between hypertonic and hypotonic. These scales refer to the etiology of each pathology, but not to their consequences or any exhibited compensatory behavior.

## Key Points

**Question** Does a 2-dimensional continuous approach provide a valid organization scheme for voice pathologies?

**Findings** In this survey study including 16 laryngologists and 23 speech-language pathologists, a scheme that arranged voice disorders on a 2-dimensional plane that was based on 2 continuous scales of organicity and tonicity was developed and evaluated.

**Meaning** In contrast with previously used categorical organizational schemes for voice disorders that demonstrated inconsistencies and categorical overlaps, this suggested scheme is a potentially simple, continuous, and dynamic approach for arranging voice disorders.

ior. The 2 scales serve as the axes of a 2-dimensional plane on which all voice pathologies may be arranged.

The concept of using a continuum (ie, spectrum) for describing physical or medical phenomena is not new, dating at least as far back as Sir Isaac Newton in the 17th century.<sup>10</sup> Since then, many disciplines have gradually shifted to using continua in conceptualizing different phenomena or conditions. They are now commonly used in a range of fields, from political orientation<sup>11</sup> and religious diversity<sup>12</sup> to gender and sexual orientation.<sup>13</sup> For example, this conceptual shift has led to the transition from using the term *autism* to *autism spectrum disorder*,<sup>14,15</sup> or viewing mental disorders, such as anxiety or depression, as positioned on a continuum rather than as dichotomous diagnoses.<sup>16,17</sup> We suggest that viewing voice disorders categorically limits flexibility in theoretical conceptualization and does not provide a sufficiently valid description of the field. Thus, this study is a preliminary attempt to present an alternative approach for arranging voice disorders using a continuous 2-dimensional approach.

## Methods

After receiving the approval of the Tel-Aviv University institutional review board, 45 highly experienced and well-established professionals (certified laryngologists or speech-language pathologists) were initially approached and offered the opportunity to participate. Because of the preliminary nature of this study, they were selected as a convenience sample. Of them, 39 (86.7%) responded, provided written informed consent, and participated in the study (16 laryngologists [41.0%] and 23 speech-language pathologists [59.0%] specializing in voice disorders). The participants were all practicing clinicians and prominent members of local and international professional laryngology and/or voice associations (eg, The Voice Foundation, International Association of Communication Sciences and Disorders, and CoMeT) who were from Austria, Brazil, China, England, France, Hong Kong, Israel, Italy, United Arab Emirates, and the US with whom the authors had previous professional interaction. Twenty-seven (69.2%) were women and 12 (30.8%) were men. Eighteen (46.2%) of the experts had

more than 30 years of experience, 7 (17.9%) had 21 to 30 years, 12 (30.8%) had 11 to 20 years, and 2 (5.1%) had 10 years or fewer.

### Survey Instrument

The survey was delivered via an online platform (Qualtrics XM). Thirty-five laryngeal and voice pathologies/conditions were taken from the Classification Manual for Voice Disorders.<sup>18</sup> These were presented to each of the experts, who rated them on 2 separate 0 to 10 rating scales: organicity and tonicity. On the organicity scale, 0 represented nonorganic and 10 organic. On the tonicity scale, 0 represented hypotonic and 10 hypertonic. The order of presentation of the 2 scales as well as the order of the different pathologies were modified randomly between raters to reduce a possible order effect. The experts were instructed to evaluate the etiology or nature of each pathology/condition rather than its possible outcome or any compensatory behavior the patient might present. All experts completed the survey in English, and all of them responded to all items. The instructions are presented in the eAppendix in the Supplement.

### Statistical Analyses

Initial analyses were performed with SAS/STAT, version 9.4 (SAS Institute). To evaluate internal consistency of ratings, a Cronbach  $\alpha$  coefficient was calculated for each scale. Interrater reliability was assessed with the intraclass correlation coefficient 1 (ICC1).<sup>16</sup> In addition, to test for interrater agreement, the  $r_{wg}$  coefficient<sup>17</sup> was calculated for each pathology. While the coefficient  $\alpha$  reflects the correlation between ratings of different judges across different pathologies,  $r_{wg}$  tests the extent to which they assign the same rating to each pathology. Mean ratings and standard deviations were calculated for each scale and for each pathology. A Pearson correlation coefficient was calculated between the 2 scales. Finally, using the Mplus 8.6 program (Muthén & Muthén), a latent profile analysis was performed to determine possible classes (clusters) of pathologies with similar values on the 2 scales, fitting models with 1 to 5 latent classes. Following recommended procedures for determining the number of classes,<sup>19</sup> we relied on the interpretability considerations, low value of log likelihood statistic, high value of entropy index, smallest value of bayesian information criteria, and a significant bootstrap likelihood ratio test. We also preferred models with a substantial proportion of cases in the smallest class based on estimated posterior probabilities. Statistical significance was set at  $P < .05$ .

## Results

Ratings of the pathologies were highly reliable, with a Cronbach  $\alpha$  of 0.98 and ICC1 of 0.60 for organicity and an  $\alpha$  of 0.97 and ICC1 of 0.49 for tonicity. Table 1<sup>20</sup> demonstrates the variation of the interrater agreement between pathologies/conditions, with an average  $r_{wg}$  value of 0.51 for organicity and 0.68 for tonicity. While interrater agreement on organicity was moderate to very strong ( $r_{wg} \geq 0.50$ ) for most pathologies, it was

**Table 1. Interrater Agreement Coefficients ( $r_{wg}$ ) and Distribution of Organicity and Tonicity Ratings**

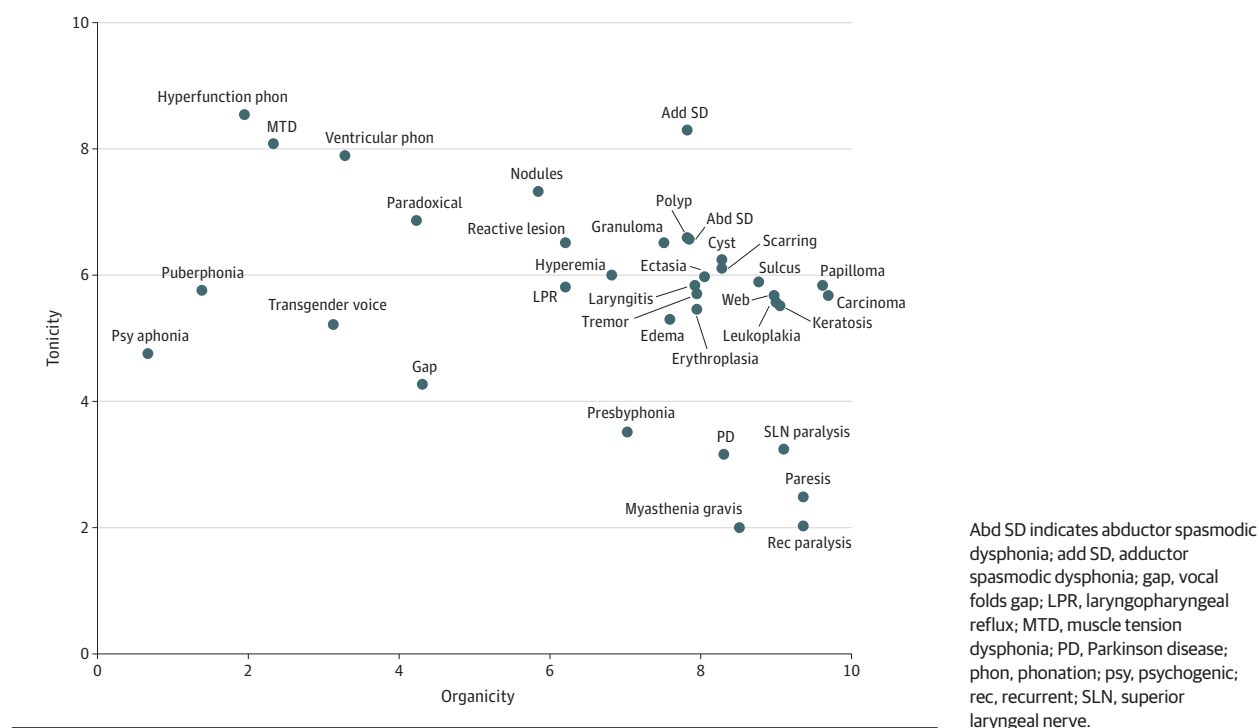
Pathology/condition	$r_{wg}$ , Mean (SD) <sup>a</sup>	
	Organicity	Tonicity
Paradoxical vocal folds movement	0 (4.23-3.22)	0.64 (6.87-1.89)
Vocal nodules	0 (5.85-3.24)	0.76 (7.32-1.55)
Vocal tremor	0 (7.95-3.19)	0.63 (5.70-1.93)
Abductor spasmodic dysphonia	0.02 (7.85-3.12)	0.19 (6.57-2.85)
Transgender voice therapy	0.05 (3.13-3.08)	0.85 (5.22-1.23)
Adductor spasmodic dysphonia	0.18 (7.82-2.86)	0.76 (8.30-1.56)
Reactive vocal fold lesion	0.25 (6.21-2.75)	0.75 (6.51-1.57)
Myasthenia gravis	0.28 (8.51-2.68)	0.71 (2.00-1.72)
Parkinson disease	0.29 (8.31-2.67)	0.46 (3.16-2.33)
Presbyphonia	0.30 (7.03-2.65)	0.56 (3.51-2.09)
Ventricular phonation	0.32 (3.28-2.62)	0.65 (7.89-1.87)
Laryngopharyngeal reflux	0.37 (6.21-2.50)	0.81 (5.81-1.37)
Muscle tension dysphonia	0.41 (2.33-2.42)	0.63 (8.08-1.92)
Ectasia	0.52 (8.05-2.20)	0.85 (5.97-1.21)
Granuloma	0.52 (7.51-2.19)	0.61 (6.51-1.97)
Polyp	0.52 (7.82-2.19)	0.74 (6.60-1.61)
Vocal folds gap	0.54 (4.31-2.15)	0.76 (4.27-1.54)
Edema	0.55 (7.59-2.11)	0.57 (5.30-2.08)
Hyperfunction phonation	0.56 (1.95-2.09)	0.78 (8.54-1.48)
Cyst	0.61 (8.28-1.99)	0.76 (6.24-1.53)
Erythroplasia	0.63 (7.95-1.93)	0.83 (5.46-1.30)
Hyperemia	0.63 (6.82-1.93)	0.87 (6.00-1.15)
Scarring of vocal folds	0.63 (8.28-1.92)	0.70 (6.11-1.74)
Laryngitis	0.64 (7.92-1.90)	0.77 (5.84-1.52)
Puberphonia	0.71 (1.39-1.70)	0.53 (5.76-2.17)
Superior laryngeal nerve paralysis	0.73 (9.10-1.64)	0.67 (3.24-1.82)
Sulcus	0.74 (8.77-1.61)	0.67 (5.89-1.82)
Glottic web	0.74 (8.97-1.60)	0.82 (5.68-1.33)
Psychogenic aphonia	0.77 (0.67-1.53)	0.20 (4.76-2.82)
Leukoplakia	0.81 (9.00-1.40)	0.81 (5.57-1.39)
Recurrent nerve paralysis	0.82 (9.36-1.35)	0.62 (2.03-1.95)
Keratosis	0.83 (9.05-1.32)	0.87 (5.51-1.12)
Vocal folds paresis	0.89 (9.36-1.04)	0.59 (2.49-2.02)
Papilloma	0.93 (9.62-0.85)	0.75 (5.84-1.57)
Carcinoma	0.95 (9.69-0.73)	0.73 (5.68-1.63)

<sup>a</sup> The  $r_{wg}$  coefficient ranges between 0 and 1. Its values are interpreted as follows: 0 to 0.30 indicates a lack of agreement, 0.31 to 0.50 weak agreement, 0.51 to 0.70 moderate agreement, 0.71 to 0.90 strong agreement, and 0.91 to 1.00 very strong agreement.<sup>20</sup>

weak or lower for 13 (37.1%). Interrater agreement on tonicity was strong for most pathologies and weak for only 3 (8.6%).

The distribution of the scores obtained for the 2 scales (means and SDs) is also presented in Table 1. Data show that the different pathologies were spread across most of the full range of both scales rather than being constrained to either end. Accordingly, all combinations of the scores on the 2 scales were evident. For example, while adductor spasmodic dysphonia was characterized as high on organicity (7.82) and high on tonicity (8.30), recurrent laryngeal nerve paralysis was characterized as high on organicity (9.36) but low on tonicity (2.03). Substantial differences were observed in the SDs of both scales,

**Figure. Distribution of the Voice Pathologies on the Organicity and Tonicity Continuous Scales Arranged as a 2-Dimensional Plane**



**Table 2. Comparison of Latent Profile Models**

No. of classes	Log likelihood	BIC	P value of BLRT	Entropy	Proportion of the smallest class
1	-148.43	311.08	NA	NA	NA
2	-135.71	296.31	.001	0.98	0.23
3	-131.19	297.94	.33	0.95	0.16
4	-123.59	293.40	.25	0.98	0.11
5	-118.51	293.90	.41	0.98	0.09

Abbreviations: BIC, bayesian information criteria; BLRT, bootstrap likelihood ratio test; NA, not available.

with smaller SDs corresponding to higher levels of interrater agreement. The correlation between the scores obtained on the 2 scales was moderate and negative ( $r = -0.38$ ; 95% CI,  $-0.63$  to  $-0.06$ ), suggesting their divergent validity.

The Figure illustrates the distribution of the experts' ratings on the 2 scales as arranged on a 2-dimensional plane. The raters' mean responses were scattered across the range of the organicity scale and slightly less so for the tonicity scale.

While the data suggested that the voice pathologies were scattered on all areas of the constructed 2-dimensional plane, the Figure suggests that they could also be viewed as arranged in 4 clusters that roughly correspond with the 4 quadrants of the 2-dimensional plane. To examine this possibility, a latent profile analysis was performed. Results for the alternative 1- to 5-cluster models are presented in Table 2.

According to the criteria of a low log-likelihood and bayesian information criteria values, the 4- or 5-cluster solutions should be preferred. The bootstrap likelihood ratio test values suggested that the 2-, 3-, 4-, and 5-cluster solutions were not significantly different. The entropy criterion favored the 2-, 4-, and 5-cluster solutions. The proportion of smallest class

in the 5-cluster solution was less than 10% (ie, included only 3 pathologies/conditions, which were considered too small). Based on these statistical findings, and the fact that the 4-cluster solution corresponded to the clusters visually prominent in the Figure, this solution was selected.

The resulting arrangement of the pathologies into the 4 clusters based on their organicity and tonicity scores is summarized in Table 3. Cluster 1 (high organicity-high tonicity) comprised 21 pathologies, cluster 2 (low organicity-high tonicity) 4 pathologies, cluster 3 (low organicity-moderate to low tonicity) 4 pathologies, and cluster 4 (high organicity-low tonicity) 6 pathologies.

## Discussion

This study introduced a new approach for arranging voice pathologies using 2 continuous scales (organicity and tonicity) rather than any of the currently available categorization schemes.<sup>2-5</sup> The results suggest that, despite the fact that many researchers and clinicians are educated to view the field of voice

Table 3. Arrangement of the Pathologies/Conditions Into 4 Clusters

Cluster	Pathologies in cluster	Organicity		Tonicity	
		Mean	$r_{wg}^a$	Mean	$r_{wg}^a$
1	Abductor spasmodic dysphonia; adductor spasmodic dysphonia; carcinoma; cyst; ectasia; edema; erythroplasia; hyperemia; keratosis; laryngitis; leukoplakia; laryngopharyngeal reflux; vocal nodules; papilloma; polyp; reactive lesion; scarring; sulcus; vocal tremor; glottal web	7.96	0.53	6.11	0.73
2	Hyperfunction phonation; muscle tension dysphonia; paradoxical vocal fold movement; ventricular phonation	2.95	0.32	7.84	0.68
3	Glottal gap; psychogenic aphonia; puberphonia; transgender voice therapy	2.37	0.52	5.00	0.59
4	Myasthenia gravis; vocal folds paresis; Parkinson disease; presbyphonia; recurrent laryngeal nerve paralysis; superior laryngeal nerve paralysis	8.61	0.50	2.74	0.68

<sup>a</sup> Interrater agreement coefficient.

and its disorders as arranged into a limited set of categories, when given the possibility of using continuous scales, highly reliable findings are provided by a group of international experts, who confirmed the continuous nature of both scales, as well as their divergent validity. The use of continuous scales for arranging voice disorders is pertinent to the theoretical and clinical conceptualization of the professional field,<sup>1</sup> as it is not limited by the definitions associated with any pathology because of that pathology's assignment to a specific category. For example, the laryngeal diagnoses of carcinomas, cysts, and polyps on vocal folds are traditionally described as organic, whereas vocal nodules are considered to have either organic or functional etiology or a combination of both. However, this study's approach places these different diagnoses on a continuum of organicity, suggesting that experts rate, for example, carcinoma as having a stronger organic component than other pathologies. In contrast, the same experts rated vocal nodules as having a lesser organic component. The fact that this approach does not necessitate separating the pathologies into distinct categories allows for such comparisons; therefore, it can potentially promote a more flexible and dynamic approach to theoretical and clinical thinking in the field of laryngology and voice disorders.

While the use of a categorical approach for arranging laryngeal pathologies/conditions has been dominant for many years,<sup>2-5</sup> it is hindered by several constraints. First, the categorical approach does not provide a naturalistic representation of the field, but rather a simplified one that can be sometimes misleading and even erroneous.<sup>21</sup> Moreover, the fact that a specific pathology could be placed in more than a single category, or that some categories are not mutually exclusive, is usually viewed as an unavoidable pitfall.<sup>9</sup> For example, as noted previously, carcinoma of the vocal folds could be in the same category as a cyst and a polyp (eg, organic or mass lesion) while their etiology, clinical manifestation, and ramifications vary considerably. Therefore, the use of a categorical approach sometimes masks the natural distribution of the pathologies, as it might overlook interpathology differences between diagnoses that are regarded as nested within a single category. Furthermore, such an approach does not reflect the complex nature of many pathologies that might arise from or be affected by organic and behavioral factors. From a theoretical perspective, the categorical approach not only fails to provide a valid representation of the field, but it also reduces statistical power, as it is based on data reduction procedures.

This has led theoreticians to advocate for refraining from using a dichotomous approach altogether and favor continuous approaches.<sup>21</sup> While a categorical approach provides a simplified model that may improve agreement, a continuous approach enhances validity<sup>22</sup> and provides a more realistic or naturalistic representation of the true nature of the observed phenomenon.

Further support for the potential use of the 2-dimensional scheme was attained from the fact that the voice pathologies were spread across (or close to) the full range of both scales such that all areas (quadrants) of the 2-dimensional plane were occupied. The data also suggested that the use of the 2-dimensional scheme appeared to reflect the emergence of 4 clusters of voice pathologies, as visualized in the Figure and presented statistically in Table 3. It should be clarified that these clusters are not to be confused with traditionally used categories. Instead, they may be viewed as representing similarities in the converged association of organicity and tonicity between pathologies with varied etiologies. This scheme places pathologies between those ranked high and low on organicity and simultaneously between hypotonicity to hypertonicity. For example, cluster 1 (high organicity-high tonicity) consisted of more pathologies compared with the other clusters. This suggests that most diagnoses in the field of voice disorders have a stronger organic component and are characterized by hypertonicity. Nevertheless, while such an interpretation may be viewed as simpler and useful in gaining additional insights on the nature of various voice pathologies, it should be stressed that the continuous nature of this model implies that clustering pathologies should be only viewed as a secondary layer of the data analysis. Therefore, this study suggests that the continuous organization of the pathologies provides a valid and more naturalistic representation compared with traditional categorical schemes.

Beyond the advantages of arranging the gamut of voice pathologies on a 2-dimensional plane, the study findings highlight a critical issue in the theoretical and clinical conceptualization of specific voice pathologies/conditions. As shown in Table 1, ratings on the organicity scale showed high interrater agreement for most diagnoses. However, some diagnoses had markedly low interrater agreement. For example, the  $r_{wg}$  coefficient value obtained on the organicity scale for vocal nodules was 0, suggesting that all possible values of the scale were used by the different experts. Inspection of the individual ratings reveals that 11 experts (28%) rated vocal nodules as low on or-



ganicity (0-3 on the scale), 18 (48%) as high on organicity (7-10 on the scale), and 10 (25%) as intermediate or having similar degrees of organic and nonorganic factors. This observation demonstrates that different professionals regard some pathologies differently and may attribute them to different etiologies. This seeming diversity could be partially explained by differences in the theoretical and clinical thinking of the different experts. However, it could be also attributed to the novelty of using the continuous scales, which might have been a challenging task for the experts, especially those accustomed to using the traditional categorical approach. Furthermore, this could also be attributed to the nature of these pathologies, which combines organic and nonorganic origins. Because of the preliminary nature and novelty of the continuous approach, resolving these discrepancies is beyond the scope of this study. Thus, future research that can replicate these findings could illuminate the specific cases in which interrater agreement is relatively low and possible differences between ratings assigned by experts from different disciplines or by professionals with varied degrees of professional expertise or experience.

### Limitations

Two limitations of this study should be considered. First, it bases the new scheme on the judgements of experts and not on objective or absolute criteria. This is because, to our knowledge, there are presently no absolute measures available for defining or for rating pathologies objectively. Thus, it could be argued that future modifications of this scheme might be required to consider alternative constructs as more knowledge accumulates on the etiology or nature of specific pathologies or as interrater agreement increases or decreases on specific

pathologies as experts become more familiar with the concept of evaluating pathologies on a continuum rather than categorically. To the extent that further refinements might be called for, we view this possibility as an advantage of this approach, as it allows for future modifications and adjustments, thus making it a dynamic, rather than a static, model.

Second, as noted previously, appropriate use of continuous scales could increase validity, but it might also reduce interrater agreement. Therefore, while this 2-dimensional scheme potentially provides a valid and reliable representation of the differences and similarities between voice pathologies, clinicians might wish to retain the use of a categorical approach to a certain degree, as it provides a more simplified view and therefore might complement the continuous scheme. Thus, future research should examine the differences between the alternative approaches for arranging voice disorders and consider their applicability for theoretical and clinical purposes.

### Conclusions

This survey study introduced a potential new approach for arranging voice pathologies using a continuous 2-dimensional plane. The results suggest the continuous nature of this scheme as well as the divergent validity of the 2 scales. The new scheme potentially provides a proof of concept for a valid and reliable new approach to arranging voice disorders beyond specific etiologies. Future research could examine how confirmed diagnoses are consistent with theoretical ratings of specific pathologies on the 2 scales.

### ARTICLE INFORMATION

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