Voice disorder: case definition in epidemiological studies

Distúrbio de voz: definição de caso em estudos epidemiológicos

Trastorno de la voz: definición de caso en estudios epidemiológicos

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Abstract

Case definition is of crucial importance in epidemiological studies. A good case definition must identify all individuals that have the problem and remove all individuals who don't have it, therefore having good sensitivity and specificity. Voice disorder is a condition difficult to measure as a result from a complex interaction of biological, psychic and social factors. As a dynamic and functional expression, the disease cannot be defined in opposition to health, but as a part of the same process. This study aimed to analyze the definition of voice disorder concept through the presence of changes in the perceptual auditory analysis of voice and visual perceptual of larynx, as well as for tests concerning the self-reference of vocal symptoms and the impact of the voice disorder handicap for the individual. The research was conducted for case definition in case-control study, with the population composed by teachers from the municipal network of São Paulo. All individuals were submitted to voice and laryngeal assessments, and they answered the Condition of Vocal Production - Teacher (CPV-P) and Voice Handicap Index (VHI) questionnaires. The results indicate that the sample was divided, in a similar way, in four different groups with respect to voice disorder both by the presence of changes in the speech language therapy and otorhinolaryngological assessments, by the reference of symptoms, and as according to the impact caused by a disorder in the social and professional life.

Keywords: *Voice Disorders; Speech, Language and Hearing Sciences; Epidemiologic Methods; Epidemiologic Studies; Measures of Association, Exposure, Risk or Outcome*

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Resumo

A definição de caso é de crucial importância em estudos epidemiológicos. Uma boa definição de caso deve identificar todos aqueles que têm o problema e excluir os que não o têm, tendo, portanto, boa sensibilidade e especificidade. Distúrbio de voz é um quadro de difícil mensuração por ser resultado de complexa interação fatores biológicos, psíquicos e sociais. Por ser manifestação dinâmica e funcional, a doença não pode ser definida em oposição à saúde, e sim, como parte de um mesmo processo. Este estudo teve o objetivo de analisar a definição do conceito de distúrbio de voz por meio da presença de alterações nos exames perceptivo-auditivo da voz e perceptivo-visual da laringe, da autorreferência de sintomas vocais e do impacto da desvantagem do distúrbio de voz para o sujeito. A pesquisa foi realizada para definição de caso em estudo caso-controle, sendo a população composta por professoras da rede municipal de São Paulo. Todas se submeteram à avaliação de voz, de laringe, e responderam questionários Condição de Produção Vocal do Professor (CPV-P) e Índice de Desvantagem Vocal (IDV). Os resultados apontam que a amostra dividiu-se, de forma similar, em quatro grupos diferenciados em relação ao distúrbio de voz, pela presença de alteração nas avaliações fonoaudiológica e otorrinolaringológica, pela referência de sintomas, e em função do impacto causado pelo distúrbio na vida social e profissional.

Palavras-chave: *Distúrbios da Voz; Fonoaudiologia; Métodos Epidemiológicos; Estudos Epidemiológicos; Medidas de Associação, Exposição, Risco ou Desfecho.*

Resumen

La definición de caso es de vital importancia en los estudios epidemiológicos. Una buena definición de caso debe identificar aquellos que tienen el problema y eliminar todas las personas que no tienen el problema proporcionando así buena sensibilidad y especificidad. Trastorno de la voz es lo resultado de una compleja interacción de factores biológicos, psíquicos y sociales. Como una expresión dinámica y funcional, la enfermedad no puede definirse en oposición a la salud, sino como parte del mismo proceso. El objetivo de este estudio fue analizar la definición del concepto de trastorno de la voz a través de la presencia de cambios en el análisis auditivo perceptivo de voz y visual perceptual de laringe, además de pruebas de auto-referencia de los síntomas vocales y del impacto de la desventaja para el individuo. La investigación fue conducida por la definición del caso en estudio caso-control, con una población compuesta por los profesores de la red municipal de São Paulo. Todos los individuos fueron sometidos a evaluaciones de la voz y la laringe, y contestaron los cuestionarios de Condición de Producción Vocal - Profesor (CPV-P) y del Índice de Discapacidad Vocal (VHI). Los resultados indican que la muestra fue dividida, de manera similar, en cuatro grupos diferentes con respecto al trastorno de la voz, por la presencia de cambios en evaluaciones profesionales de logopedia y otorrinolaringología, por la referencia de los síntomas, y según el impacto causado por un trastorno de la voz en la vida social y profesional.

Palabras clave: *Trastornos de la Voz; Fonoaudiología; Métodos Epidemiológicos; Estudios Epidemiológicos; Medidas de Asociación, Exposición, Riesgo o Desenlace*

Introduction

Case definition is a specific set of criteria that an individual must meet to be classified as a "case" in an investigation[1]. This definition includes both clinical and laboratory criteria, such as epidemiological criteria, which are related to the individual, time and space. That is, this is how the disease identification evaluation will be standardized to enable the comparison of data and prevent the occurrence of bias in a research.

In this sense, how to define a voice disorder case for epidemiological studies?

Conceptually, dysphonia is any change that would prevent, hinder or hamper voice production[2]. It's a symptom that composes the voice disorder framework and manifests itself through different auditory and visual signals. The concept involves difficulty in voice emission of any degree and origin and, although it may seem to be simple and consensual, it becomes complex since it originates from the definition of normality of the voice.

As a functional disease of multiple and complex causation, the voice disorder is not restricted to the sum of its factors, making it difficult to precisely define case in researches. If on the one hand the complaint may be a sufficient condition for the therapeutic action in clinical practice, regardless of the presence of auditory or visual signals, on the other hand there is a need for standardization of the case definition in researches.

The diseases defined by the prefix *dis* have a functional design and are characterized by the experience of being sick[3]. This means that, in the presence of a voice disorder, "not all healthy individuals are free of disease and not all individuals free of disease are healthy"[4]. The voice disorder affects individuals who might not be considered "sick", since they are physically and socially active, while others have significant limitations, both personal and professional.

In the absence of standardization, the case definition of voice disorder takes different forms and classifications in the studies. Most studies of prevalence[5,6,7,8,9] are based on self-reported symptoms in questionnaires, while a few make use of a professional evaluation[10,11,12,13,14,15,16,17,18,19]. However, this can be not so specific to define the disease under discussion as there is a wide variation in the results depending on how the symptoms are measured. The prevalence of fatigue when speaking on teachers, for instance, ranges from 18% to 88%, which suggests a need to set limits on symptoms frequency and duration to improve the methodological precision for definition of the real prevalence rate of vocal disorder[19]. The use of symptoms reference is more appropriate for a screening, provided that only current symptoms are included, which have an impact on the daily lives of the individuals[20].

On the other hand, there is growing trend to the use of self-evaluation protocols with respect to the impact of the vocal disorder for the individual[21]. The International Classification of Functioning, Disability and Health (ICF), of the World Health Organization[22], defines as a handicap how the individual adapts to the environment due to his disability or incapacity. The concept of incapacity is defined in the negative sense of functionality, resulting from the interaction between the disorder presented by the individual (organic or structural), the limitation of his activities and the restriction on social integration, with environmental factors as facilitators or barriers[23]. Under this perspective, the WHO recommends that health evaluations include not only indicators of changes in the frequency and severity of the disease according to the evaluation of professionals, but also aspects that reveal the social conditions, of welfare and quality of life of individuals[24].

In the face of such recommendations, it is considered that it is impossible to define case in voice disorder in opposition to the absence of any symptom or signal. Similarly, this definition should not be made strictly by results of vocal and laryngeal evaluations, under the risk of disregarding early or insidious forms of the disease, when symptoms may be present without some organic signal[20], as well as disregarding the impact of this problem for each individual. There is, therefore, the challenge to define precise criteria for such definition.

The **objective** is to analyze the definition of the case concept in voice disorders through the presence of changes in the perceptual auditory analysis of voice and visual perceptual of larynx, as well as test concerning the self-reference of symptoms and the impact of the voice disorder handicap for the individual.

Methods

Observational, cross-sectional study. The population was composed of 352 teachers from the municipal network of São Paulo. Only female participants were included because they represent the large majority in the studied population, in addition to present a higher prevalence of vocal disorder when compared to male teachers[11]. We excluded teachers who showed changes in vocal folds that were not associated with the use of voice[2] and who were on medical leave, functional readjustment or performing administrative functions, since in these cases the use of the voice differs from the teaching activities.

Research approved by the Research Ethics Committees of the Faculty of Public Health - University of São Paulo (FSP-USP) under no. 173/07, Hospital do Servidor Público Municipal de São Paulo (HSPM-SP) under no.101/07, which was conducted from July 2007 to May 2009 for case definition in a case-control study[25]. The teachers who participated received clarifications and agreed to participate in the study by signing the Free and Informed Consent Form.

All individuals were submitted to speech-language and otorhinolaryngological evaluations, and they answered the Condition of Vocal Production - Teacher (CVP-T)[26] and Voice Handicap Index (VHI) questionnaires[24].

The speech samples were collected in acoustic cabins and evaluated in a simultaneous analysis of three judges with experience in voice. The vocal quality was ranked using the GRBASI scale [27] in with changes (level 2 or 3) and without changes (level 1 or 2), considering that most of the teachers has vocal changes, even though many have mild changes. The videolaryngoscopies were performed by the same otorhinolaryngologist, soon after evaluation, and the participants were considered with changes in the presence of injury, irritating or structural change or change of coaptation of vocal folds and no change in the absence of such changes.

The CVP-T questionnaire identified characteristics of vocal use and symptoms reported, classifying as with symptoms (sometimes, always) and without symptoms (never, rarely). The VHI quantified the impact of the handicap caused by the voice disorder. A Cohen's Kappa test was performed to evaluate the agreement between the speech-language and otorhinolaryngological diagnostics; a Chi-square test was performed to determine the association between symptoms and diagnostic groups; and a Kruskal–Wallis one-way analysis of variance was performed to compare the averages of the VHI.

Results

The VHI scale presented excellent internal consistency (Cronbach α VHIG-0.93, VHIE-0.93, VHIF-0.91, VHIO-0.94).

Most of the teachers are between 30-49 years (72.6%), are married (58.8%), have higher education course (92.7%), are titular professor (95.8%), work in a school (52.8%), have 15 years of profession on average, teach more than 20 hours/week (71.0%), don't smoke (88.6%), drink rarely or never (82.6%). The vocal and laryngeal evaluations showed a good agreement level (76.6%, Kappa=0.52).

After a joint analysis of the evaluations, the participant was classified into four groups: **non-case**, no changes in both evaluations (29.4%); **case 1**, changes in the vocal evaluation and no changes in the visual perceptual evaluation (7.9%); **case 2**, no changes in the vocal evaluation and changes in the visual perceptual evaluation (15.5%); **case 3**, changes in both evaluations (47.2%).

Table 1 shows the distribution of four groups of individuals according to the reference of vocal and non-vocal symptoms. It's possible to notice the similarity of the values found among groups **case 1** (changes in vocal assessment and no changes in the otorhinolaryngological evaluation) and **case 3** (changes in both evaluations), which may indicate cases of early voice disorder, although without organic expression. The **case 2** group presents intermediate values between the other two groups, **case 1 and case 3**.

Table 2 shows the distribution of four groups of individuals according to general and partial scores of the Vocal Handicap Index (VHI). These results are similar to those in Table 1, where the **non-case** group has lower indexes, the values of **case 1 and case 3** groups have greater differences and the **case 2** group has intermediate values.

Table 1. Distribution of four groups of individuals according to the reference of vocal and non-vocal symptoms.

Vocal and non-vocal symptoms	non-case		Case 1		Case 2		Case 3		P value (c2)
	no.	%	no.	%	no.	%	no.	%	
Hoarseness									
no	50	49.0	2	7.4	12	21.8	11	6.6	<0.001
yes	52	51.0	25	92.6	43	78.2	156	93.4	
Loss of voice									
no	82	79.6	12	44.4	26	50.0	70	42.4	<0.001
yes	21	20.4	15	55.6	95	57.6	95	57.6	
Failing voice									
no	61	60.4	3	11.1	15	28.3	35	21.1	<0.001
yes	40	39.6	24	88.9	38	71.7	131	78.9	
Shortness of breath when talking									
no	75	72.1	14	53.8	28	51.9	72	43.6	<0.001
yes	29	27.9	12	46.2	26	48.1	93	56.4	
Rough voice									
no	73	71.6	10	38.5	23	46.0	58	35.6	<0.001
yes	29	28.4	16	61.5	27	54.0	105	64.4	
Voice change									
no	85	84.2	11	40.7	28	54.9	92	56.4	<0.001
yes	16	15.8	16	59.3	23	45.1	71	43.6	
Pain when speaking									
no	68	66.7	6	22.2	27	50.0	63	38.2	<0.001
yes	34	33.3	21	77.8	27	50.0	102	61.8	
Fatigue when speaking									
no	51	50.0	3	11.5	14	25.5	22	13.3	<0.001
yes	51	50.0	23	88.5	41	74.5	144	86.7	
Effort when speaking									
no	49	47.6	2	7.4	11	20.4	23	13.9	<0.001
yes	54	52.4	25	92.6	43	79.6	143	86.1	
Total	105	100	26	100	54	100	167	100	

Table 2. Distribution of four groups of individuals according to general and partial scores of the Vocal Handicap Index (VHI).

VHI	Non-case (n=105)	Case 1 (n=27)	Case 2 (n=55)	Case 3 (n=167)	P value*
Functional	16.67	31.20	22.64	32.93	<0.001
Emotional	13.10	35.37	22.91	32.49	<0.001
Organic	26.55	53.89	42.86	57.05	<0.001
General	18.77	40.15	29.47	40.82	<0.001

*p: Kruskal–Wallis one-way analysis of variance

Discussion

Case definition is of crucial importance in epidemiological studies. A good case definition must identify all individuals that have the problem and remove all individuals who don't have it, therefore having good sensitivity and specificity.[1]. Voice disorder is a condition difficult to measure as a result from a complex interaction of biological, psychic and social factors. As a dynamic and functional expression, the disease cannot be defined in opposition to health, but as a part of the same process.

The speech-language and otorhinolaryngological evaluations are complementary in understanding this disorder and the authors[27,28,29] propose an association of more than one method in order to

have precise knowledge of the vocal dynamic and of the larynx conditions. In this study, the voice perceptual-auditory and laryngeal perceptual-visual evaluations presented good agreement level. There was no consensus in groups **case 1** (7.9%), participants with changes in vocal evaluation and no changes in otorhinolaryngological evaluation, and **case 2** (15%), no change in the vocal evaluation and with changes in the perceptual-visual evaluation. Two possibilities are considered in the case 1. The first is the existence of minimal structural changes in vocal folds which can't be observed during a videolaryngoscopy, since approximately 30% of these changes are diagnosed only in a surgery[33]. The other possibility is that this is an initial vocal expression, without the corresponding organic signal. Case 2 may have teachers who, even with some changes in vocal folds, have a well adapted voice[2].

With respect to the self-reference of vocal symptoms, the form used by most studies to identify the presence of a voice disorder, the results of this study indicate that any symptom, alone, is sufficiently specific to distinguish individuals who are sick of those who aren't. This can be confirmed by observing that 52% of the participants of the non-case group reported to currently have hoarseness, 50%, fatigue when speaking, and 54%, effort when speaking. However, the absence of some symptoms in the non-case group, such as loss of voice (79.6%), shortness of breath when speaking (72.1%) or voice change (84.2%), may be a predictive indicator of the absence of a voice disorder. On the other hand, the presence of hoarseness (93.4%), fatigue when speaking (86.7%) and effort when speaking (86.1%) in the case 3 group suggest the presence of this disorder. If there are changes in the otorhinolaryngological evaluation without impact on vocal quality, it can be assumed a remission stage of the vocal disorder through the vocal emission more adapted to the biological condition.

The results comparison of the Vocal Handicap Index (VHI) with the four groups reinforces this hypothesis, when it indicate that the case 1 group presents scores that are close to those of the case 3 group, while the case 2 group shows greater indexes (Table 2), revealing a greater handicap when there is an impairment of vocal quality. Considering that the same problem can affect each individual differently[23], the better the adaptation in the light of the dysfunction (organic and/or structural), the lower the limitation of activities and restriction in social participation of the individual, with the environmental factors as facilitators or barriers[22]. Under this perspective, individuals who have worse vocal quality indicate greater impact in their personal and professional relations[24].

Conclusion

The results of this study indicate that the sample was divided, in a similar way, in four different groups in relation to the voice disorder, both from organic perspective, by the presence of changes in speech-language and otorhinolaryngological evaluations, and with respect to the complaint, by symptoms reference, as according to the impact caused to the individual.

As a functional disease of multiple and complex causation, the voice disorder is not restricted to the sum of its factors, making it difficult to precisely define case in researches. If on the one hand the complaint may be a sufficient condition for the therapeutic action in clinical practice, regardless of the presence of vocal or laryngeal signals, on the other hand there is a need for standardization the case definition in researches.

The diseases defined by the prefix *dis* have a functional design and are characterized by the experience of being sick. This means that, even presenting a voice disorder, individuals might not be considered "sick", since they are physically and socially active, while others have significant limitations, both personal and professional.

Therefore to estimate the real magnitude of this disease, the importance of evaluating the impact of the problem for the functional, social and professional life of the individual should be highlighted, as much as identifying the symptoms or conduct a voice and larynx evaluation.

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