

Case Report

Relato de Caso

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Descritores

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Achados de neurorradiologia e voz no acidente vascular encefálico

ABSTRACT

Neurological dysphonias are vocal disorders followed by injuries or changes in the nervous system. Stroke is the second leading cause of death worldwide and the brain damage caused by it can affect communication in several aspects. The changes in the vocal features caused by these lesions are poorly described regarding the location and extent of cerebral involvement. The aim of this study was to describe vocal parameters of ten elderly patients affected by stroke according to the topography of the lesion at neuroimaging. We recorded from them: perceptual and auditory evaluation of voice and laryngeal diadochokinesis. Neuroimaging studies were classified according to location, extent, laterality territory of brain injury. The results show a population of extensive middle cerebral artery strokes and strokes with varied location in the brain. The subjects' voices showed predominantly the presence of roughness, breathiness, richness and instability, and change in laryngeal motor control, demonstrated by reduced speed and instability in laryngeal diadochokinesis. These features presented in patients with extensive middle cerebral artery stroke and in patients with short strokes with varied location in the brain. The results suggest that the vocal features in the assessed cases do not depend only on the topography of the brain damage. Thus, it is important to consider the patient as unique in clinical evaluation.

RESUMO

As disfonias neurológicas são distúrbios vocais que acompanham lesões ou alterações no sistema nervoso. O acidente vascular encefálico (AVE) é a segunda causa de morte no mundo e os danos cerebrais causados podem afetar a comunicação do indivíduo em diversos aspectos. As alterações de voz características dessas lesões são pouco descritas quanto à localização e extensão do acometimento cerebral. O objetivo deste estudo foi descrever as características fonatórias de dez idosos acometidos por AVE, de acordo com a topografia da lesão observada ao exame de neuroimagem. Todos os participantes tiveram amostras de voz gravadas e foi realizada avaliação perceptivo-auditiva da voz e da diadococinesia laríngea. Os exames de neuroimagem foram classificados quanto à localização, extensão, lateralidade e território de vascularização da lesão cerebral. Os resultados mostram uma população de AVEs extensos de artéria cerebral média e AVEs de localização variada no cérebro. As vozes dos sujeitos mostraram predominantemente presença de rugosidade, sopro, instabilidade e pastosidade, além de alteração no controle motor laríngeo, demonstrado pela velocidade reduzida e instabilidade na repetição de vogais. Essas características ocorreram tanto no grupo de pacientes com AVE extenso de lesão da artéria cerebral média como em pacientes com AVEs menores de localização variada no cérebro. Os achados sugerem que as manifestações vocais dos casos estudados não dependem apenas da topografia da lesão. Daí a importância de considerar as individualidades de cada paciente no momento da avaliação clínica.

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INTRODUCTION

The newer methods of clinical research, such as structural and functional neuroimaging, as well as theoretical advances, make us ponder the concept of lesion–symptom relationships on language and speech. Thus, when considering the neural circuits responsible for language and speech, a lesion in the region of these circuits tends to affect the function as a whole, although in varying degrees and forms⁽¹⁾.

Vocal disorders resulting from neurological lesions may be present in cases of dysarthria, which is the abnormal speech due to neurological injury and affects the execution of the motor action. In dysarthria, changes occur in one or more of the following components: breathing, phonation, articulation, resonance, and prosody⁽²⁾. Some authors have described the following common symptoms in dysarthria: pneumophonoarticulatory incoordination, impaired vocal quality, changes in pitch and loudness, hypernasality, and changes in the articulation pattern⁽³⁾.

There are few studies that relate the changes in phonation with neuroradiological images obtained in patients with neurological disorders. The evaluation of voice, especially in cases of stroke, is also poorly described, given its importance in the restoration of the communication functions of these patients.

In general, studies in this field observed the changes in patients during the acute phase of the onset, with the vocal and speech assessments conducted within the first 72 hours⁽³⁻⁵⁾, but little is known about the changes in the stable phase of the onset. It is noteworthy that these studies evaluated the auditory-perception of the quality of patients' voice and oral motor control through diadochokinesia (DDK) tasks. This assessment reflects the adequacy of the individual's neuromotor maturation and integration, considering a test of neurological skills in which the subject must perform fast repetitions of the same syllable or vowel. In the case of the vowel, only the laryngeal motor control ability is evaluated; an understudied aspect⁽⁶⁾, but it provides important information.

Among the studies that observed the area of injury in the brain in cases after a stroke, one of them⁽³⁾ reported that the lesions responsible for dysarthria were located in the supratentorial region in 45.6% of the cases, and in the infratentorial region in 54.4% of the cases; the supratentorial strokes were found more often on the left (74.2%) than on the right (25.8%) hemispheres. The same group of researchers, in another study⁽⁴⁾, reports that dysarthria in extracerebellar stroke was most often caused by lesions in the left cerebral hemisphere and its intensity was more pronounced when the lesion was in the same hemisphere, regardless of its topography.

Changes such as weakness, slowness, or incoordination of the laryngeal muscles may be identified by voice. Furthermore, the identification of laryngeal signs and symptoms can facilitate the early differential diagnosis of certain diseases⁽⁷⁾. In more severe cases of neurological disorders, examination of phonatory characteristics can reveal important information about the basic neuropathology. Aiming to

a better understanding of the interference of changes in the central nervous system (CNS) in voice emission and the pursuit of more substrate for rehabilitation, the study of phonatory characteristics in relation to the location and extent of injury is justified.

Thus, the aim of this study was to describe the phonatory characteristics of 10 patients affected by stroke according to the topography of the lesion, observed by neuroimaging exams.

PRESENTATION OF CLINICAL CASES

This study was approved by the Research Ethics Committee (under protocol no. 139/2010), and the subjects signed a free and informed consent form.

Ten subjects with clinical diagnosis and who underwent imaging tests that confirmed brain injury from stroke were evaluated. The sample consisted of individuals aged over 60 years, affected by stroke, confirmed by computed tomography (CT). Six subjects were female and four were male, and all of them were affected by stroke between 6 months and 3 years and 8 months before the time of the evaluation, with an average of 2 years and 1 month after the occurrence of ischemia. The average age for women was 77.3 years and that for men was 73 years.

A laryngeal nasoendoscopy was performed to exclude individuals who had laryngeal lesions not related to the stroke, such as polyps, nodules, cysts, leukoplakia, and sulcus vocalis.

The interpretation of CT scans of the skull was performed by two neuroradiologists, aiming to characterize brain lesions regarding their location, area of vascular irrigation, extension, and laterality. Strokes with more than 3 cm in length and affecting more than one cerebral lobe were considered extensive.

Auditory-perceptual voice was assessed with Consensus Auditory-Perceptual Evaluation of Voice (CAPE-V), an instrument developed by a group of speech-language pathologists and voice specialists who are part of the Special Interest Division 3 — Voice as Voice Disorder of American Speech-Language and Hearing Association (SID-3 – ASHA).

CAPE-V assesses six predetermined parameters in three tasks: sustained vowel, specific sentences from a protocol, and spontaneous speech. To indicate the degree of deviation observed, we used a linear analog scale, with 100 mm for each parameter. Evaluators indicated the degree of perception from normal to the deviation, for each parameter of the scale. The parameters analyzed in the protocol were the following: overall degree of change, roughness, breathiness, strain, pitch, and loudness. This protocol allows the inclusion of two extra parameters and, in this study, the additional parameters used were instability in the emission of sustained vowel and richness in the two tests involving speech.

The voices of 10 patients are part of a group of samples recorded in a preliminary study, which were presented randomly to three judges with expertise in auditory-perceptual voice analysis. The reliability of intra- and inter-evaluators was verified using the Kappa statistical test; results ranged from satisfactory to almost perfect reliability. For the description of the results

of this sample, the average value assigned by the three judges in the visual analog scale was considered for each parameter.

The DDK, which is the ability to perform rapid repetitions of relatively simple patterns comprising oppositional contractions, was evaluated by repeating two distinct vowels separately, /a/ and /i/, uninterruptedly.

The analysis of the emissions of the vowels /a/ and /i/ was performed by the Motor Speech Profile – Advanced (MSP) software from KayPENTAX. The parameters of the DDK are provided automatically by the MSP software and are shown in Chart 1.

Data analysis was performed in a descriptive manner in relation to all subjects, as well as in relation to two groups of subjects according to the location and extent of injury: one with strokes with extensive lesions of the middle cerebral artery and the other with smaller-sized strokes with lesions in varied locations of the brain.

RESULTS

In the analysis of skull CT scans, a large homogeneous group of strokes (example shown in Figure 1) were found, and in five patients (three women and two men), the middle cerebral artery was found affected. These are indicated in the

tables, differentiating them from the rest of the group, which is formed by smaller strokes of varying locations in the brain (example shown in Figure 2). In addition, other findings of the examination have been described: ventriculomegaly, which corresponds to an increase in the size of the brain ventricles without increased intracranial pressure; calcifications in the basal ganglia; mega cisterna magna, characterized by an increase and morphological alteration of the cisterna magna; and congenital alteration. Such findings were observed in the two groups (Table 1).

All subjects in the sample had some degree of breathiness and roughness in their voice, instability in the emission of sustained vowels, and richness during speech; at least half of the patients showed some degree of vocal strain (Table 2). The laryngeal DDK showed slowness and instability in emissions (Table 3). These findings were present regardless of the extent of brain injury.

DISCUSSION

In analyzing the results of the auditory-perceptual evaluation, we see generally high values of parameters in all subjects in the sample, especially with the sustained /a/ test and independent phrases test, regardless of the type of stroke.

Chart 1. Parameters analyzed in the laryngeal diadochokinesia

Parameters	Unit	Notes
DDKmT	Mean of the DDK rate	/s Number of vocalizations per second, which represents the DDK speed
DDKdpP	Standard deviation of the period of DDK	ms
DDKcvp	Coefficient of variation of the period of DDK	% Measures the degree of variation of the rate in the period, indicating the ability to keep a rate of constant vocalizations
DDKJitP	Disturbances of the period of DDK	% Measures the degree of cycle to cycle variation in the period, indicating the ability to keep a rate of constant vocalizations
DDKcvi	Coefficient of variation of the intensity peak of DDK	% Measures the degree of variation of each vocalization, indicating the ability to keep the intensity of vocalizations

Caption: DDK = diadochokinesia

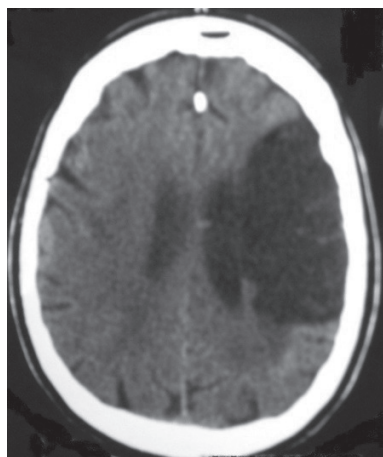


Figure 1. Computed tomography showing an extensive stroke of the middle cerebral artery, affecting more than one cerebral lobe

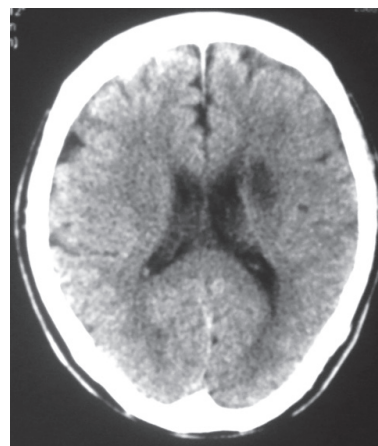


Figure 2. Computed tomography showing a stroke smaller than 3 cm, affecting the insula

Table 1. Characterization of the sample according to gender, age, time of brain affection in the evaluation and characteristics of the lesion observed on computed tomography

Case	Gender	Age (years)	Time of stroke	Brain hemisphere	Lobe	Extension	Lesion of the middle cerebral artery	Other
1	F	82	3.8 years	L	FITP	>	Yes	V
2	F	81	11 months	R	FTP	>	Yes	V
3	F	80	3 years	R	FITP	>	Yes	–
4	M	80	1.7 years	R	FT	>	Yes	–
5	M	70	2.6 years	R	FP	>	Yes	–
6	F	62	8 months	L	Insular	<	No	–
7	F	87	3 years	R	F	<	No	V
8	F	72	6 months	R	F	>	No	MCM
9	M	66	3 years	R	O	>	No	CBG
10	M	76	2 years	L	I	<	No	–

Caption: F = female; M = male; L = left; R = right; F = frontal; I = insular; T = temporal; P = parietal; O = occipital; Extension: > (greater) or < (smaller) than 3 cm; Other test findings: V = ventriculomegaly; MCM = mega cisterna magna; CBG = calcification of the basal ganglia; (–) absent

Table 2. Average values of the parameters of the Consensus Auditory-Perceptual Evaluation of Voice protocol evaluated by the judges for each test and for each subject

Subjects	Gender/Age	Extensive lesions with affection of the MCA					Smaller lesions without affection of the MCA				
		1	2	3	4	5	6	7	8	9	10
		F/82	F/81	F/80	M/80	M/70	F/62	F/87	F/72	M/66	M/76
Tests	Parameters										
Sustained /a/	Overall degree	50.00	60.67	33.00	22.33	34.67	38.33	59.33	26.00	36.67	48.33
	Roughness	28.67	51.00	28.67	25.67	26.00	7.00	25.00	2.67	32.00	31.33
	Breathiness	17.33	16.00	14.67	0.00	0.00	9.00	49.67	23.67	15.67	41.00
	Strain	0.00	24.00	0.00	8.67	31.00	0.00	22.00	0.00	4.33	0.00
	Pitch	0.00	8.67	7.33	0.00	20.00	0.00	3.33	0.00	21.00	0.00
	Loudness	0.00	20.00	7.33	0.00	0.00	3.00	30.67	8.00	0.00	21.33
Phrases	Instability	15.67	30.33	23.00	12.33	15.00	35.67	13.00	19.33	22.00	17.33
	Overall degree	70.33	24.67	39.00	18.00	33.67	54.67	70.67	15.67	26.33	55.33
	Roughness	66.67	14.33	31.33	12.67	28.67	35.00	65.67	11.33	25.00	42.33
	Breathiness	6.33	0.00	0.00	0.00	17.67	0.00	20.67	6.67	0.00	33.67
	Strain	15.00	13.67	18.33	0.00	24.00	0.00	8.00	0.00	0.00	39.00
	Pitch	24.00	8.67	2.00	0.00	14.00	0.00	11.67	0.00	0.00	6.33
	Loudness	11.67	0.00	0.00	0.00	0.00	18.00	4.00	3.33	0.00	4.33
Spontaneous conversation	Richness	25.33	0.00	22.67	2.33	5.67	27.33	31.33	6.33	15.33	19.67
	Overall degree	–	20.67	64.00	23.67	–	26.33	58.67	21.00	31.33	64.67
	Roughness	–	14.33	59.67	26.67	–	26.67	55.00	7.33	22.67	58.33
	Breathiness	–	0.00	9.33	3.33	–	0.00	19.00	7.33	4.00	27.67
	Strain	–	21.33	12.33	0.00	–	0.00	21.00	0.00	0.00	27.00
	Pitch	–	5.33	15.67	10.00	–	20.67	8.33	0.00	6.33	0.00
	Loudness	–	0.00	9.67	0.00	–	0.00	3.67	9.00	0.00	18.00
Richness	–	0.00	25.00	0.00	–	10.00	40.00	5.00	13.67	34.67	

Caption: MCD = middle cerebral artery; F = female; M = male; (–) refers to the nonevaluation of the parameter, due to difficulty or nonexecution of the task requested by the participant

One study⁽⁸⁾ showed that acoustic measurements indicating the presence of noise were more pronounced in patients affected by stroke than those in healthy patients, suggesting greater tendency for noisy voices, with a greater degree of vocal deviation in these subjects. Regarding the visual analog scale in voice assessment, the literature refers to the indicated value of 34 voice deviation points as the threshold for considering that the individual failed the voice screening⁽⁹⁾. Although the cutoff values for each of the parameters tested in the CAPE-V were not indicated, it was possible to notice a uniform roughness in the voice of the subjects in the sample, in all tests, as well as

the choice of the items instability and richness in most cases, which is noteworthy.

In the sustained /a/ test, all subjects evaluated presented some degree of instability in the voice, which is characterized by oscillation of any frequency and intensity, regardless of pace. Urban et al.⁽⁴⁾, when describing the vocal quality of the participants in their study, reported vocal instability.

Regarding richness, only one subject was considered to lack this parameter in the evaluation by the three judges; the remainder of the sample presented values ranging from 6 to 31 mm in the sentences test and 5–40 mm in spontaneous

Table 3. Laryngeal diadochokinesia values provided by the Motor Speech Profile – Advanced software, by KayPENTAX, for each subject

		Extensive lesion with affection of the MCA					Smaller lesion without affection of the MCA				
Subjects		1	2	3	4	5	6	7	8	9	10
Gender/Age		F/82	F/81	F/80	M/80	M/70	F/62	F/87	F/72	M/66	M/76
Parameters	Unit										
A	mT (/s)	3.00	3.67	4.25	2.30	2.19	2.20	3.02	2.30	3.33	2.37
	dpP (ms)	29.07	169.0	23.36	21.02	98.82	65.39	72.90	21.02	49.54	28.51
	cvP (%)	8.73	61.95	9.94	4.83	21.69	14.42	22.00	4.83	16.49	6.77
	JitP (%)	1.77	11.30	3.50	1.67	16.65	10.54	9.04	1.67	6.94	3.65
	cvI (%)	2.57	5.05	1.69	3.68	1.59	1.78	3.59	3.68	3.05	2.23
I	mT (/s)	2.80	1.57	3.30	2.49	2.17	2.37	2.78	2.49	3.13	2.62
	dpP (ms)	24.36	15.26	32.46	77.94	63.42	149.0	1.82	77.94	89.21	63.94
	cvP (%)	6.83	2.39	10.72	19.37	2	5	6	19.37	27.88	16.75
	JitP (%)	2.24	1.04	4.68	6.87	F/62	F/87	F/72	6.87	5.92	8.28
	cvI (%)	3.03	3.38	1.66	2.53	2.19	2.20	3.02	2.53	1.25	3.15

Caption: MCA = middle cerebral artery; F = female; M = male; mT = mean of the diadochokinesia rate; dpP = standard deviation of the period; cvP = coefficient of variation of the period; JitP = disturbance of the period; cvI = coefficient of variation of the intensity peak

conversation. Some authors reported that richness in voice reflects slowness, imprecise articulation, and hypernasality, characteristics that may be present in neurological disorders such as stroke^(4,10), due to loss of muscle tone^(10,11). Interestingly, subjects who showed absence of this parameter presented extensive strokes of the middle cerebral artery in the right cerebral hemisphere, but we can relate this good performance with other studies^(4,5) that showed the evolution in the speech of these patients after the acute phase of the onset.

The literature suggests the DDK task to assess motor coordination and speed of movement of the of the phonoarticulatory organs in patients with neurological disorders^(4,11).

Some authors⁽¹²⁾ reported that normal individuals possess appropriate adjustments in laryngeal mechanisms that enable rapid glottal opening and closure during production of the consonants and vowels of syllables. This adjustment can be modified in cases with neurological disorders.

Dividing individuals from the sample into two groups, extensive strokes in the middle cerebral artery and smaller strokes in varied locations, through the average, it is possible to observe that subjects with extensive strokes in the middle cerebral artery tend to have higher DDK speed, both of /a/ and of /i/, also a smaller standard deviation of DDK in both tests, a higher coefficient of variation of the DDK period in the DDK of /a/ and equated to the other group in the DDK of /i/ lower values of disruption for the DDK period (JitP) in both tests and higher values relate to the coefficient of variation of the peak intensity of the DDK (cvI), also in the two tests.

Magalhães⁽¹³⁾ evaluated the DDK of elderly individuals aged 70–79 years. Although we know that it is not possible to correlate the data of the elderly individuals in this study because there is no statistical analysis of the results, and considering that these parameters are poorly studied, it is observed that the rate of the DDK is similar in healthy elderly patients and those who had stroke. However, even if a statistical comparison is not possible, it is possible to describe that, regarding instability parameters (cvP, JitP,

and cvI), women affected by stroke showed higher values than those in the study cited, both for the DDK of /a/ and of /i/. Men affected by stroke showed higher values than healthy elderly only for the DpP and cvP parameters of the DDK of /i/; in the rest of the tests, the values in both populations are quite similar. Therefore, the women in this study showed difficulty in maintaining constant vocalizations, both in period and in cycle to cycle, and a difficulty in maintaining the intensity of vocalizations when compared to the elderly population between 70 and 79 years without neurological changes. When analyzing case by case, we can say that only subjects 2 and 4 had a good performance on the laryngeal DDK task, close to healthy elderly⁽¹³⁾ in all parameters; the remaining subjects showed slower DDK and greater instability in period, cycle to cycle, and relate to intensity.

Data in the literature indicate that the laryngeal DDK is slower in the case of neurological changes⁽⁶⁾. Few authors have addressed the assessment of oral DDK in post-stroke patients^(4,11) and only one of them evaluated the laryngeal DDK in this population⁽¹¹⁾, having found a DDK rate varying between 0.75 and 4.75 emissions per second.

When we look jointly to all vocal assessments carried out, we can see that, regardless of the location and extent of brain injury, the subjects who achieved good performance tended to show that in all of the evaluations, as especially shown by cases 2, 4, 6, and 9. Subjects who had a worse performance also tended to have difficulty in all tests, which was observed mainly in subjects 1, 7, and 10.

There are no reports that compare the types of voices to the area of brain injury. However, both the perceptual findings on vocal quality and the perceptual aspects of oral DDK are described in the literature relate to the types of dysarthria, and these can be defined according to the area of injury in the CNS.

Spastic dysarthrias result from upper motor neuron injury and are extensively described in stroke cases. There is also a separate description, made by some authors, of unilateral

upper motor neuron dysarthria, in which 90% of the causes are unilateral strokes in this brain region. Flaccid dysarthrias, resulting from injuries between the brainstem and the neuromuscular junctions, lead to muscle weakness, paresis, or paralysis that may affect orofacial muscles or laryngeal structures, which makes voice and speech weak. Hypokinetic dysarthrias in stroke cases are commonly associated with bilateral ischemia in the thalamic region. These cases can be termed as vascular parkinsonism. Hyperkinetic dysarthrias, not very common in stroke cases (about 1% of this type of dysarthria are due to some vascular injury), appear when there is brainstem stroke, which leads to movement disorders such as chorea, dystonia, and essential tremor. Mixed dysarthrias, also common in this population, arise from various types of injuries, as there is a combination of one or more types of dysarthria⁽¹⁴⁾.

Some patients in this sample, with slight vocal changes and absence of changes in other speech components, did not fall into the category of diagnosis of dysarthria. No cases of ataxic dysarthria were observed, because no participant showed cerebellar lesion. Moreover, no lesions in the brainstem were observed in the tests, which rules out cases of hyperkinesia.

Other patients in this sample fit more easily in the description of spastic dysarthria or unilateral upper motor neuron dysarthria, whose phonatory characteristics are lowered pitch, roughness, strained voice, frequency breaks, short sentences, and reduced speed in tests such as the DDK. Another possible description is mixed dysarthria, wherein speech characteristics from more than one type of dysarthria occur, common in patients with multiple strokes. Owing to the vocal characteristics of this sample, such as weakness or richness in speech and articulatory imprecision, some cases could be described as spastic dysarthria and flaccid characteristics.

It is important to consider that this study shows that lesions in similar areas showed distinct vocal expressions; therefore, the most basic classification of dysarthrias⁽²⁾ does not always cover all aspects to be described. Due to brain injuries resulting from stroke being quite variable, which may affect the brain in different locations and sizes, classifying neurological dysphonia in these cases is complex. According to Aronson⁽¹⁵⁾, the vocal manifestations presented by patients in this study fall in relatively constant neurological disorders of voice.

Factors such as neuronal plasticity, early intervention, overall health data, and the aging process are directly related to rehabilitation and patient performance. Therefore, one may suggest that vocal changes in brain injury will not always be specific for each type of injury, because the recovery of the functions observed in the stable phase of onset and common vocal deterioration in aging are often associated. In addition, decline in cognitive ability and apathy observed in the elderly with stroke can also affect speech and, consequently, the vocal aspects of this patient.

This study reported the characteristics of the vocal behavior of some individuals after a stroke in a stable phase of the onset, some with extensive lesion and the involvement of

the middle cerebral artery and others with smaller lesions in varied locations. Although case reports present limitations due to the small number of participants and the inability to create multiple groups with more specific brain lesions, as there is no similar report of such vocal expressions in the literature, the presentation of cases contributed to the characterization of the voice of these and individuals, and to the information that the voice quality and laryngeal motor control do not always have specific characteristics according to the affection of the brain.

Studies involving a large number of post-stroke patients and individuals of the same age without neurological diseases, using instruments for auditory-perceptual evaluation, laryngeal motor control, and other procedures, should be undertaken to provide more data for a better understanding of vocal manifestations in this population.

FINAL COMMENTS

Vocal characteristics found in all subjects were roughness, breathiness, sometimes strain, instability (during the emission of sustained vowels), and richness (during speech). The presence of changes in laryngeal motor control was observed, as evidenced by the slowness and instability in emissions of laryngeal DDK. These features occurred both in patients with extensive stroke lesion of the middle cerebral artery and in patients with minor strokes in varying locations in the brain.

**JFG participated in the data collection and drafting of the article; AGB participated as a co-advisor to the study, supervising all collection procedures, actively participating in the conception and design of the study and drafting of the article; GBF actively participated in the conception and design of the study and drafting of the article; AYF participated as an advisor to the study, also aiding in the conception and design of the study, drafting of the article and interpretation of imaging tests.*

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