

Phonotherapeutic Intervention in Patients With Mucosal Leishmaniasis Sequelae

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Summary: Purpose. To characterize the voice before and after speech-language intervention, with Humming nasal sound in patients with sequelae Mucosal Leishmaniasis (ML) and Cutaneous Leishmaniasis (CL).

Methods. Collection of phonation /a:/ from 44 patients with ML and CL for perceptual voice analysis and computed acoustic. The Wilcoxon nonparametric test and Fisher's exact test were used, with significance level of 5%.

Results. It was observed, prespeech therapy, that 27.7% of participants with ML presented asthenic vocal quality, and for the acoustics characteristics there was a statistically significant result for measures of frequency, frequency disturbance, noise, and subharmonic measurements, indicating phonatory instability, weakness, and noise emission giving the emission a feeling of vocal weakness. After therapy, the subharmonic segment measurements for the group with ML, showing reduction noise emission. Patients with CL had more grade 1 instability (36.4%), indicating tremor in vocal tract structures. After speech therapy, this group presented a reduction in the degree of roughness and reduction of the frequency disturbance measures, indicating a decrease in tension in the larynx and pharynx.

Conclusion. Even after completing treatment for LM, patients may experience vocal changes due to the sequelae of the disease, like vocal alterations due to nasal lesions or in other locations that interfere in the correct vocal emission. As for participants with CL, no vocal changes would be expected, since these patients present thorax, leg and arm lesions that would not cause problems for the voice. Nevertheless, the two groups of participants presented vocal changes to different degrees before vocal therapy. However, it was observed that patients with ML present vocal alterations with more severe degrees. After the speech-language intervention, the participants of both groups showed vocal improvement, but the group with CL presented more vocal benefits, possibly due to the previous vocal alterations not being so severe.

Key Words: Voice—Mucosal Leishmaniasis—Cutaneous Leishmaniasis—Voice disorders.

INTRODUCTION

Leishmaniasis is an infectious parasitic disease of great concern to public health. Brazilian Northeast suffers a significant incidence of this disease, mainly American Tegumentary Leishmaniasis, although it is possible to find it from the south of the United States to the north of Argentina.^{1–4}

This disease is characterized for having several clinical presentations and can cause disfiguring and disabling lesions with severe psychosocial consequences. There are three classical types of clinical presentation: Mucosal Leishmaniasis (ML), Cutaneous Leishmaniasis (CL) and Disseminated Leishmaniasis.^{2,5}

ML causes destructive lesions of the upper respiratory tract which can cause from a small perforation of the nasal septum to the destruction and collapse of the entire nasal pyramid. Approximately 5% patients healed from CL, develop ML after months or years.^{5–9} Some of the symptoms of this pathology are: nasal obstruction, cough, rhinorrhea and epistaxis. However, these symptoms and the complaints reported by the patients such as dysphagia, odynophagia, dysphonia, dyspnea, depend on the area affected by the LM (mouth, nose, pharynx, and larynx).^{10,11}

Dysphonia, one of the complaints reported by those patients, can significantly impact their quality of life due to communication impairment, which, consequently, affects social and professional relationships.^{12,13} In addition, there is a psychological factor,¹⁴ that can further worsen the quality of life and voice of those patients.¹⁵

Voice rehabilitation of patients with ML can rely on voice therapy programs that significantly benefit communication^{16,17} such as semioccluded vocal tract exercises indicated either in

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cases of voice disorders or in cases of individuals with normal voice but, who pursue voice economy and efficiency.^{18–20}

These exercises are performed by occluding the anterior region of the vocal tract, promoting retroflex resonance toward the vocal folds,¹⁷ which generates increased phonatory comfort and vocal resistance, in addition to reducing laryngeal tension.^{21,22}

An example of these exercises is the nasal sound “humming,” which provokes phonation harmony. This vocal technique ensures adequate interaction between source and filter, minimizing the impact suffered by the vocal folds during emission. With this, the risk of lesions is reduced and resonance and vocal projection are improved.²³ These effects are only possible because of the energy dissipated toward the supralaryngeal cavities. In this way, the vibration generated in the facial tissues will allow lighter phonation due to the relaxation of laryngeal, pharyngeal, and oral structures.^{24,25}

Thus, we intend to verify and compare the auditory perceptual and acoustic vocal characteristics before and after the speech-language intervention, with Humming nasal sound, in patients with sequelae of ML and CL.

Method and patients

Study design

A cross-sectional analytical study, of experimental nature was conducted at the Dr Jackson Lemos Costa Health Clinic located in the village of Corte de Pedra, in the municipality of Tancredo Neves in southeastern Bahia, from February 2016 to May 2017. Patients were divided into two groups (Participants with sequelae of CL) and (Participants with sequelae of ML), with the purpose of characterizing their voices and verifying the vocal changes occurred after the application of vocal technique. To this end, the technique was applied in only one session, where three series of nasal sound were performed, collecting the voice of the participants before and after the technique.

All participants signed an informed free consent term – Termo de Consentimento Livre e Esclarecido or CF (TCLE; acronym in Portuguese) in accordance to the ethical principles established in Norm 466/12, of the National Committee of Ethics in Research and approved by the Committee of Ethics in Research of COMHUPES-UFBA, under number 43/2015.

Patient selection

All participants selected for the study, according to inclusion and exclusion criteria, had already completed the treatment for Leishmaniasis. After the speech-language evaluations, the participants who presented vocal alterations were referred for vocal treatment, and the patients had the choice to continue the treatment.

Participants were selected according to the following inclusion criteria: aged from 18 years, aiming at avoiding the alterations of the voice change period,^{26,27} to 70 years to include older male patients who are more frequently affected by

ML;^{28,29} participants who had ML and CL, and were diagnosed by parasitology, histology, culture, immunology (provided by the Immunology Service – SIM- of HUPES), and otorhinolaryngology; with concluded medical treatment for leishmaniasis; with sequelae of nasal, buccal, pharyngeal and laryngeal lesions diagnosed by a physician, with or without voice complaints; who signed the TCLE.

As for exclusion criteria, patients with the following characteristics were excluded from the study: smokers, alcohol consumers;^{27,30} those who did not finish the drug treatment for ML or CL, those with history of voice therapy, history of Diffuse, Disseminated or Mucocutaneous (concomitant) Leishmaniasis, those with history of respiratory diseases (bronchitis, asthma, allergic rhinitis, sinusitis); who had the flu on the day of the evaluation; who presented a history of neurological problems;^{27,30} who reported other granulomatous diseases (paracoccidioidomycosis, squamous cell carcinoma, basal cell carcinoma, lymphomas, rhinophyma, rhinosporidiosis, entomophthoromycosis, Wegener granulomatosis, Wirchowian leprosy, tertiary syphilis, tuberculosis, sarcoidosis, and cocaine-induced midline granuloma); who had other laryngeal pathologies (minimal structural changes in the coverage and organic lesions);³⁰ who had traumatic septal perforation or by drug use diagnosed by a physician.³¹

For the performance of exclusion and inclusion criteria it was done anamnesis, including questions on complaints of voice disorders, aspects that could interfere in voice performance or in the execution of the evaluations, use of larynx aggressive agents that could favor the emergence of laryngeal disorders, respiratory diseases, date of American Tegumentary Leishmaniasis (ATL) diagnosis and voice conditioning through speech-therapy techniques and otorhinolaryngological evaluation, including oroscopy, rhinoscopy, and videolarngoscopy were carried out targeting the identification of laryngeal or other disorders in accordance to the exclusion criteria; and evaluation of the Stomatognathic System of CL patients, who can present facial deformities, observing the posture, mobility and tonicity of these structures

Sample

All participants in this study were enrolled in a post-treatment follow-up list of Leishmaniasis. Patients were called for review via telephone call. At the time of the medical appointment, participants were informed about the research and questioned about their interest in participating in the study.

The sample was composed of 63 participants interested in the research. Participants were numbered and randomized, using the simple random sampling process until reaching a minimum of 44 cases. After being subjected to the inclusion and exclusion criteria, two participants with sequelae of CL were excluded because they did not complete all the examinations and one of them because he presented a nodular lesion on the right vocal fold (Figure 1). On the other hand, only one participant with sequelae of ML was excluded because he presented a granulomatous disease (paracoccidioidomycosis) (Figure 1), and was referred to monitoring and specific treatment.

Laryngoscopy Characteristics		
	Mucosal Leishmaniasis	Cutaneous Leishmaniasis
N(%)		
Mobility	22(100%)	22 (100%)
Symmetry	22 (100%)	22 (100%)
Complete Glottic Coaptation	21(95,45%)	19(86,36%)
Incomplete Glottic Coaptation		
Irregular Slit	1 (4,54%)	2 (9,09%)
Triangular posterior Slit		1 (4,54%)
Presence of Mucosal wave	22 (100%)	22 (100%)
Supraglottic Activity	0 (0%)	0 (0%)
Participantes excluídos		
Vocal nodules		X
Paracoccidioidomycosis	X	

FIGURE 1. Laryngoscopic characteristics of patients with leishmaniasis sequelae.

Thus, four participants were excluded, being necessary to continue the lottery until reaching the n determined by sample size calculation. Therefore, according to sample size calculation assuming a level of significance of 5% (α), a sample power of 80% ($1-\beta$), and a simple random sampling process for the statistical analyses, sample size was 22 participants per group, totaling 44 cases.

Data collection

To collect vowel /a:/ emission, patients were asked to be in orthostatic position and to emit the sound in a sustained manner in usual pitch and loudness, at maximum phonation time and without using the respiratory reserve.^{27,30,32} Emission was recorded in a digital *Zoom Model Q3 Handy Recorder* professional voice and video recorder, with PCM audio format, quantization of 16 bits, capture frequency of 96 kHz, keeping a mouth-recorder distance of 4 cm and 90°, in a quiet environment.^{27,30,32}

Collection was carried out immediately before and after the complete execution of the Nasal Sound vocal technique, in just one session, performed by the participants in maximum phonation time, in three series of 15 repetitions,^{27,33,34} with a 30-second rest between series, with the patients sitting and in complete silence.^{27,35} The technique was demonstrated and monitored by a speech therapist to avoid incorrect performance.^{27,36,37}

To perform the technique, the participants closed their lips and clenched their teeth. Gradually they opened their teeth only slightly, keeping their jaws relaxed and their tongue on the floor of the mouth. Then they emitted the prolonged “hum” sound, imagining emitting the sound of

the vowel / u /, with perception of the vibration in the mask area (nose/mouth).²³ During the technique, participants were sitting with their feet flat on the floor, an upright spine with no cervical dislocation, with a 90° angle between the chin and the neck, without increasing the muscular contraction of the shoulder girdle, maintaining the constant rhythm between one repetition and another, without making use of the expiratory reserve and, avoiding the fluctuation or variability of pitch and/or loudness.^{27,33,36,38}

Ingestion of 250 ml water was allowed,³⁹ considering a possible vocal tract dryness with increased air flow. However, this did not interfere at the glottic level until the final vowel emission was collected, since water reached the larynx in a systemic way, not interfering with the results of the research.

Concerning vowel /a:/ emission, auditory perceptive evaluation was performed through the RASATI scale that evaluates hoarseness (R), level of roughness (A), breathiness (S), asthenia (A), strain (T) and instability (I), using the following rating scale: 0 = normal: when no voice alteration is perceived by the listener; 1 = slight, when the alteration is evident and 3 = for extreme voice alterations.⁴⁰ This evaluation was conducted by four speech-therapists judges, with expertise in the voice area, who did not participate as authors of this study and were blinded about the purposes of the research. The information that the evaluators received was on the age and gender of the patients, and the recordings were delivered through a file storage and sharing service based on the “cloud computing” concept with individual accounts.³⁰

To this analysis it was carried out an inter-rater agreement through the Kappa coefficient. The interpretation of

the test is as follows: values less than 0 correspond to no agreement; values between 0 and 0.19 represent poor agreement; values between 0.20 and 0.39 correspond to good agreement; values between 0.40 and 0.59 correspond to moderate agreement; values from 0.60 to 0.79 correspond to strong agreement; and values between 0.80 and 1 correspond to an almost perfect agreement.⁴¹ It was observed a strong agreement between the judges, with an index of 0.78. Only the answers of these judges were selected and grouped to the data analysis, due to a greater reliability.

Also, a computed acoustic analysis was performed, using vowel /a:/ emission and the *Multi Dimensional Voice Program Advanced* (MDVPA) from Kay Pentax, in which vocal attack was eliminated and the end of emission discarded so that these stretches did not alter signal analysis, once the ends of prolonged emissions usually present decreases of amplitude and frequency, creating therefore the interval of 5 seconds for the analysis window, which was the smallest sustain time obtained in the group.^{27,30,42}

Threshold values for males proposed by the MDVPA software^{30,33,43} were used as reference of the extracted measures. In addition, the fundamental frequency parameters were analyzed considering the following values as reference of normality: 80 Hz–150 Hz for males.⁴⁴ The measures automatically extracted by MDVPA, with sampling rate of 44 kHz and 16 bits, were the frequency measures: f0; maximum f0 (fhi); minimum f0 (flo); Standard deviation of f0 (STD); frequency perturbation measures: absolute *Jitter* (Jita); *Jitter* percentage (Jitt); pitch relative average perturbation; pitch perturbation quotient (PPQ); smoothed pitch perturbation quotient (sPPQ); variation of f0 (vf0); amplitude perturbation: *Shimmer* in dB (ShdB); *Shimmer* percentage (Shim); amplitude perturbation quotient; smoothed amplitude perturbation quotient; amplitude variation (vAm); noise measures: noise-to-harmonics ratio (NHR);

voice turbulence index (VTI); soft phonation index (SPI); voice break measures: degree of vocal breaks; number of vocal breaks; measures of voiceless or unvoiced segments: number of unvoiced segments (NUV); degree of unvoiced segments; measures of subharmonic segments: degree of subharmonic components (DSH); number of subharmonic components (NSH).^{27,30,43}

Statistical analysis

SPSS 17.0 was used for the statistical analysis, including social-demographic data and clinic characteristics. In addition, to compare de auditory measures before and after applying the vocal technique to the group with sequelae of ML and the comparison between the group with sequelae of CL and ML, Wilcoxon nonparametric test was applied having in mind that 80% of the distributions were not normal, with a level of significance of 5% ($P < 0.05$). On the other hand, Fisher exact test was used to compare the data of the RASATI scale, before and after vocal treatment between groups (ML and CL).

RESULTS

The sample was composed of 44 participants, 22 in each group, all males, with average age of 57.59 ± 6.49 for the participants with sequelae of ML and 35.13 ± 11.28 for the participants with sequelae of CL. All participants were from the rural zone, occupationally active, most of them exerting activities in the field (Table 1).

Of the 22 patients with ML, 72.72% presented lesions on the nose (Table 2), and the symptoms more reported during the illness period were nasal crusts (86.36%), septal perforation (40.90%), epistaxis (36.36%), nasal obstruction (31.81%), rhinorrhea (27.27%), anosmia (13.3%), collapse of the nasal pyramid 1 (9.09%), hyposmia (4.54%) and

TABLE 1.
Demographic Distribution of Patients With Cutaneous Leishmaniasis and Mucosa

Variables	Mucosal Leishmaniasis		Cutaneous Leishmaniasis	
	N	%	N	%
Gender				
Female	0	0	10	45,45
Male	22	100	12	54,54
Occupation				
Farmer	20	90,90	16	72,72
Bricklayer	2	9,09	0	0
Housewife	0	0	4	18,18
Domestic	0	0	1	4,54
Seamstress	0	0	1	4,54
Previous history of Leishmaniasis				
Yes	11	50	6	27,27
No	11	50	16	72,72
Age				
Age	Mean	Standard deviation	Mean	Standard deviation
	57,59	$\pm 6,49$	38,27	$\pm 12,84$

N, number of subjects. %, percentage.

TABLE 2.
Location of Lesions of Patients With Leishmaniasis Before Drug Treatment

Location of Injuries	Mucosal leishmaniasis	
	N	%
Nose	16	72,72
Nose+Mouth	1	4,54
Nose+Pharynx	3	13,63
Mouth+Pharynx	1	4,54
Nose+Pharynx+Mouth	1	4,54

N, number of subjects. %, percentage.

dysphonia (4.54%). Concerning the disease staging degree: 9.52% presented grade I, 28.57% grade II, 23.80% grade III, 28.57% grade IV and 9.52% grade V.

The comparison of the results of the RASATI scale between groups with ML and CL before the speech therapy showed statistically significant results, with 27.7% participants with ML showing the voice quality asthenia, indicating that this characteristic is more frequently associated with ML. In contrast, more patients with CL had instability grade 1 (36.4%) than patients with ML (Table 3).

After applying the nasal sound technique, participants of both groups presented reduction of the grade of hoarseness, although those of the CG presented greater reduction than those of the SG, with statistically significant results (Table 3).

Table 4 shows, that in the comparison of ML with CL before the application of the vocal technique, statistically significant results were obtained for frequency measures (fhi, STD), frequency perturbation measures (vF0), noise

measures (SPI) measures of voiceless or unvoiced segments (NUV), measures of subharmonic segments (NSH), indicating association of these parameters with the group with ML, which presented higher values than the CL group. However, the CL group presented higher SPI (noise measure) to emission than the ML group.

On the other hand, after the speech therapy intervention there was statistically significant reduction of the frequency perturbation measures (PPQ, sPPQ, vF0) for the CL group and of the measures of subharmonic segments (NSH, DSH) and of STD for the ML group.

Table 5 shows the acoustic measures of the MDVPA, before and after applying the vocal technique, only for the group ML, which presented statistically significant reduction of the noise parameters (VTI, NHR), although with increase of the SPI. Moreover, there was increase of the degree of unvoiced segments.

Figure 1 shows the laryngoscopic characteristics of patients with LM and LC sequelae.

DISCUSSION

Throughout Brazilian territory, the predominance of Leishmania (Viannia) brasiliensis⁴⁵ was observed, with a large growth since 2000, with the state of Bahia being the second, after Maranhão, to report⁴⁶ and, on the other hand, the southern region of the country presented the lowest number of cases.⁴⁷

The LM and CL are two of the main clinical forms of LTA, the former being considered simpler than the latter. Some authors have shown that 5%–10% of patients who receive treatment or cure for CL may develop ML.^{5,8}

TABLE 3.
Comparison of Auditory Perceptual Parameters Between the Mucous and Cutaneous Leishmaniasis Groups, Before and After the Vocal Technique

		Mucosal Leishmaniasis								Cutaneous Leishmaniasis								P Value
		0	1	2	3	0	1	2	3	0	1	2	3					
B E F O R	R	N	%	N	%	N	%	N	%	N	%	N	%	N	%	N	%	0,087
		6	27,3	7	31,8	4	18,2	5	22,7	11	50	6	27,3	5	22,7	0	0	
	A[†]	6	27,3	5	22,7	7	31,8	4	18,2	13	59,1	4	18,2	5	22,7	0	0	0,077
	S	19	86,4	2	9,1	1	4,5	0	0	22	100	0	0	0	0	0	0	0,223
	A	16	72,7	1	4,5	5	22,7	0	0	21	95,5	1	4,5	0	0	0	0	0,048*
	T	16	72,7	4	18,2	1	4,5	0	0	17	77,3	2	9,1	3	13,6	0	0	0,511
A F T E R	R	N	%	N	%	N	%	N	%	N	%	N	%	N	%	N	%	P value
		10	45,5	8	36,4	4	18,2	0	0	11	50	10	45,5	1	4,5	0	0	0,479
	A[†]	6	27,3	10	45,5	6	27,3	0	0	18	81,8	4	18,2	0	0	0	0	0,000*
	S	21	95,5	1	4,5	0	0	0	0	22	100	0	0	0	0	0	0	1,000
	A	17	77,3	4	18,2	1	4,5	0	0	21	95,5	1	4,5	0	0	0	0	0,185
	T	18	81,8	4	18,2	0	0	0	0	14	63,6	1	4,5	1	4,5	0	0	0,345
I	20	90,9	2	9,1	0	0	0	0	14	63,6	6	27,3	2	9,1	0	0	0,083	

Fisher's exact test. * Statistically significant values ($P < 0.05$). N, number of subjects. %, percentage. R, hoarseness; [†]A, asperity; S, thunder; A, asthenia; T, tension; I, instability.

TABLE 4.
Comparison of Vocal Technique Application Between Cutaneous and Mucous Leishmaniasis Groups Through Acoustic Means of MDVPA

		ML			CL			P Value
		Medium	Q1	Q3	Medium	Q1	Q3	
B E F O R	F0	161,35	124,65	200,02	142,57	131,99	165,91	0,330
	Fhi	205,89	152,49	225,67	147,19	135,29	185,18	0,017*
	F10	133,67	99,26	173,60	137,84	128,59	158,62	0,734
	STD	3,63	1,93	2,17	1,75	1,27	2,24	0,003*
	Jita	59,59	25,36	131,40	36,86	24,52	62,88	0,142
	Jitt	0,93	0,38	1,74	0,52	0,36	0,91	0,119
	RAP	0,56	0,21	1,06	0,28	0,20	0,56	0,124
	PPQ	0,52	0,21	0,98	0,28	0,21	0,50	0,121
	sPPQ	0,86	0,59	1,91	0,66	0,48	0,86	0,051
	vf0	1,91	0,99	5,66	1,21	0,88	1,47	0,019*
	ShdB	0,33	0,15	0,76	0,29	0,25	0,43	0,751
	Shim	3,68	1,81	8,27	3,29	2,79	4,86	0,769
	APQ	3,14	1,91	6,37	3,19	2,63	4,76	0,805
	sAPQ	6,27	3,45	8,46	6,25	4,33	8,47	0,664
	vAm	15,79	8,63	22,52	13,70	10	18,83	0,769
	NHR	0,15	0,12	0,22	0,13	0,11	0,15	0,074
	VTI	0,068	0,04	0,086	0,055	0,044	0,068	0,152
	SPI	4,62	3,09	6,26	7,41	5,75	9,46	0,002*
	DVB	0,000	0,000	0,000	0,000	0,000	0,000	0,153
	NVB	0,000	0,000	0,000	0,000	0,000	0,000	0,153
NUV	0,000	0,000	12,25	0,000	0,000	0,000	0,031*	
DUV	0,000	0,000	6,94	0,000	0,000	0,12	0,109	
DSH	0,000	0,000	1,22	0,000	0,000	0,000	0,206	
NSH	0,000	0,000	1,25	0,000	0,000	0,000	0,034*	
A F T E R	Medium	Q1	Q3	Medium	Q1	Q3	P value	
	F0	162,94	135,425	193,33	154,75	134,92	177,37	0,330
	Fhi	168,03	139,39	211,84	156,98	139,02	185,57	0,285
	F10	138,76	107,19	180,34	151,22	131,25	169,19	0,565
	STD	2,17	1,63	3,60	1,62	1,15	2,41	0,017*
	Jita	54,04	28,66	111,35	34,22	20,82	65,56	0,149
	Jitt	0,91	0,47	1,57	0,47	0,33	0,95	0,119
	RAP	0,55	0,28	0,95	0,27	0,19	0,57	0,093
	PPQ	0,65	0,21	1,06	0,28	0,19	0,53	0,022*
	sPPQ	0,69	0,56	1,64	0,55	0,49	0,81	0,050*
	vf0	1,31	1,05	2,50	1,15	0,85	1,46	0,022*
	ShdB	0,35	0,17	0,62	0,27	0,19	0,32	0,231
	Shim	3,81	1,61	6,04	3,12	2,09	3,50	0,366
	APQ	3,15	1,93	5,38	2,83	1,96	3,46	0,519
	sAPQ	5,55	3,67	7,94	5,29	4,12	6,88	0,796
	vAm	13,35	8,78	19,66	11,42	6,97	13,64	0,130
	NHR	0,13	0,12	0,15	0,13	0,12	0,14	0,372
	VTI	0,052	0,044	0,07	0,047	0,037	0,061	0,213
	SPI	6,76	4,20	10,05	6,65	5,18	10,14	0,769
	DVB	0,000	0,000	0,000	0,000	0,000	0,000	0,317
NVB	0,000	0,000	0,000	0,000	0,000	0,000	0,317	
NUV	0,000	0,000	4,25	0,000	0,000	0,000	0,081	
DUV	0,000	0,000	2,31	0,000	0,000	0,000	0,081	
DSH	0,000	0,000	1,16	0,000	0,000	0,000	0,043*	
NSH	0,000	0,000	1,20	0,000	0,000	0,000	0,018*	

Wilcoxon ratings test. Q1, first quartile; Q2, second quartile; Q3, third quartile. The data marked with (*) are statistically significant.

Abbreviations: f0, fundamental frequency; fhi, highest fundamental frequency; flo lowest fundamental frequency; STD, standard deviation of f0; Jitta, absolute jitter; RAP, média relativa da perturbação do *pitch*; PPQ: quocientede perturbação do *pitch*.; sPPQ, smoothed pitch perturbation quotient; vf0: Variação da f0;ShdB, shimmer in dB; APQ: quociente de perturbação da amplitude; sAPQ, smoothed amplitude perturbation quotient; vAm: variação da amplitude; NHR, noise to harmonic ratio; VTI, voice turbulence index; SPI, soft phonation index; DVB, degree of voice breaks; NVB, number of voice breaks; NUV, number of unvoiced segments; DUV, degree of voiceless; DSH, degree of subharmonics; NSH, number of subharmonic segments.

TABLE 5.
Acoustic Measurements of the MDVPA Before and After Vocal Technique (Nasal Sounds) in the Leishmaniasis Mucosa Group

	Before Vocal Technique			After Vocal Technique			P Value
	Medium	Q1	Q3	Medium	Q1	Q3	
F0	161,35	124,65	200,02	162,94	135,425	193,33	0,838
Fhi	205,89	152,49	225,67	168,03	139,39	211,84	0,262
F10	133,67	99,26	173,60	138,76	107,19	180,34	0,610
STD	3,63	1,93	2,17	2,17	1,63	3,60	0,176
Jita	59,59	25,36	131,40	54,04	28,66	111,35	0,424
Jitt	0,93	0,38	1,74	0,91	0,47	1,57	0,980
RAP	0,56	0,21	1,06	0,55	0,28	0,95	0,949
PPQ	0,52	0,21	0,98	0,65	0,21	1,06	0,775
sPPQ	0,86	0,59	1,91	0,69	0,56	1,64	0,321
vf0	1,91	0,99	5,66	1,31	1,05	2,50	0,187
ShdB	0,33	0,15	0,76	0,35	0,17	0,62	0,759
Shim	3,68	1,81	8,27	3,81	1,61	6,04	0,775
APQ	3,14	1,91	6,37	3,15	1,93	5,38	0,849
sAPQ	6,27	3,45	8,46	5,55	3,67	7,94	0,638
vAm	15,79	8,63	22,52	13,35	8,78	19,66	0,243
NHR	0,15	0,12	0,22	0,13	0,12	0,15	0,039*
VTI	0,068	0,04	0,086	0,052	0,044	0,07	0,024*
SPI	4,62	3,09	6,26	6,76	4,20	10,05	0,050*
DVB	0,000	0,000	0,000	0,000	0,000	0,000	0,500
NVB	0,000	0,000	0,000	0,000	0,000	0,000	0,750
NUV	0,000	0,000	12,25	0,000	0,000	4,25	0,310
DUV	0,000	0,000	6,94	0,000	0,000	2,31	0,014*
DSH	0,000	0,000	1,22	0,000	0,000	1,16	0,820
NSH	0,000	0,000	1,25	0,000	0,000	2,00	0,625

Wilcoxon ratings test. Q1, first quartile; Q2, second quartile; Q3, third quartile. The data marked with (*) are statistically significant.

Abbreviations: f0, fundamental frequency; fhi, highest fundamental frequency; flo lowest fundamental frequency; STD, standard deviation of f0; Jitta, absolute jitter; RAP, Média relativa da perturbação do *pitch*; PPQ, quociente de perturbação do *pitch*; sPPQ, smoothed pitch perturbation quotient; vf0, Variação da f0; ShdB, shimmer in dB; APQ, quociente de perturbação da amplitude; sAPQ, smoothed amplitude perturbation quotient; vAm, variação da amplitude; NHR, noise to harmonic ratio; VTI, voice turbulence index; SPI, soft phonation index; DVB, degree of voice breaks; NVB, number of voice breaks; NUV, number of unvoiced segments; DUV, degree of voiceless; DSH, degree of subharmonics; NSH, number of subharmonic segments.

The CL is a chronic and benign disease,^{48,49} being the most common form of ATL, which may not manifest itself over the years or evolve to cutaneous lesions. The characteristic CL lesions may present as single or multiple, ulcerated,^{14,50} usually painless, round or oval in appearance, with erythematous, infiltrated and firm appearance, with circumscribed and elevated borders, reddish and with coarse granulations.^{51,52} They often affect exposed areas of the body, such as limbs.^{14,50}

Usually, the appearance of ML occurs after primary skin lesion healing (2%–5% of cases), but it can also occur simultaneously to the CL frame.^{14,53,54} However, the true incidence of LM in Brazil is not known, often due to the lack of notification of these cases.⁵⁵

The mucosal lesions can be configured as destructive, potentially mutilating, reaching the upper aerodigestive pathways.^{56,57,58,59} Mucosal involvement in ATL is considered a serious manifestation of the disease, which makes it impossible for the person to work and withdraw from social interaction.¹⁴ Among men, the mucosal form is more common, besides they exhibit more septal perforation and there

is propagation of the lesions to external nasal cavity structures.²⁸

In this study, 59.09% (n = 26) participants were aged between 25 and 50 years, all were rural dwellers and most of them worked in agriculture. Studies show that there may be variation concerning the spread of cases among the different age groups, indicating the possibility of household, peridomiciliar and occupational transmission, because children under six and the elderly are generally indoors, while people of productive age have higher risk of having the disease because of their occupations in the forest, usually agriculture or extractivism (Table 1).^{28,60,61}

The sample of this study was composed of males and most studies show that males are more affected by ATL,^{10,28} that the mucosal form is more frequent and that they can develop more severe forms of the disease, with high incidence of perforations and involvement of the external structures of the nasal cavity.²⁸ This prevalence of ATL among males, may occur due to their occupational or leisure activities such as hunting, fishing or camping which are still more common among them (Table 1).^{60,62}

ML has an insidious onset and although it presents scarce initial symptomatology,⁶³ has lesions with a great destructive potential,⁶⁴ affecting the mucosa and cartilage of the upper airways and digestive tract,^{7,58,59,63,65,66} disfiguring and disabling the individuals affected by this disease.^{7,58,59,63,66} Among the symptoms frequently reported in the literature,^{10,31,64,67} this study found higher occurrence of nasal crusts, septal perforation, epistaxis, nasal obstruction, and rhinorrhea during the course of the disease (Table 2).

As a result of the symptoms of rhinorrhea, nasal crusts, epistaxis, hyposmia, septal perforation, collapse of the nasal support (tapir nose),^{9,10,68} individuals may present breathing problems, such as breathing in a forced manner.¹⁰ Also, many patients have pain when swallowing, and sialorrhea, caused by lesions on the hard and soft palate, uvula, gingiva, and pharynx,^{10,64,68} and may manifest dysphagia.⁶⁸ Moreover, mastication may be impaired because of the involvement of the gums and dental interstices, where coarse and prominent granulations may develop, reaching the upper lip, often sparing the tongue.^{51,68} All this can cause speech impairment, with phoneme distortions or articulatory difficulties.

Clearly, the most prevalent symptoms indicate the nasal region as the most affected site (72.72%) (Table 2). The literature shows that most lesions are located in the nose,^{10,31,64,67} and that the involvement of the nasal mucosa can occur without previous cutaneous disease, revealing that *Leishmania* penetration occurred at the cutaneous–mucosal transition of the nose structure.¹⁰ Moreover, it is believed that parasite metastasis can be brought to the upper airways and digestive tract through lymphatic or hematogenous route^{10,64} and unusually by direct contact of the mucosa with a cutaneous lesion as in the case of a newborn whose mother has a cutaneous lesion on the nipple and who developed a mucosal lesion in the mouth.¹⁰

In addition, some studies^{7,10,29,31,64,69} have shown that, following the nasal site, lesions may appear on the palate, pharynx, larynx, and gum. However, in the present study, it was observed that most of the lesions occurred on more than one site concomitantly (Table 2). In addition, even without the presence of lesions in the larynx, we observed lower incidence of dysphonia, which, according to some authors, may be associated with lesions on the pharynx and oral cavity.¹¹ Moreover, this symptom may also be due to presbyphonia, since the CL group is composed of participants with higher age.^{30,70,71} Even considering these results, the development of mucosal lesions is still not well understood. Several factors are related to these lesions such as, the parasite, the host, and the magnitude of the immunological response,^{10,49,64,72} as well as socioeconomic and environmental factors or even contiguity.⁶⁴

The voice of these patients may be altered due to the location of the lesions sequelae, which were present on the nose, oral cavity, larynx, and pharynx, causing changes in resonance and vocal quality.⁷³ There is generalized laryngeal inflammation particularly in the region of the pyriform sinuses. The vocal folds may be moving well, but the phonation may be weak

(asthenic) and the muscle contraction tension may be impaired by granulomatous formation and subsequent fibrosis.^{51,68}

Although no laryngeal lesions were found in this research (Figure 1), it is known that voice production depends on the harmony between breathing, phonation, and resonance which express the emotions and represent people's personality.^{74,75} Thus, when there is no harmony in that set, one of the consequences may be dysphonia,^{74,76} which will impair or alter voice emission, affecting natural voice.^{77,78}

The auditory perceptive analysis of the vocal characteristics of the groups studied evidenced a statistically significant association of asthenia grade 2 with the ML group (27.7%) and of voice instability grade 1 with the CL group (36.4%) (Table 3). Asthenia gives a feeling of weakness to emission and instability is the abnormal change of the voice.⁷⁹ In the case of asthenia, it was also found that, acoustically, the values Jitter and Shimmer were higher when compared to the CL group (Table 4), however, without statistical significance. Studies have shown that this voice quality may be related to high indices of these acoustic parameters.⁸⁰

Moreover, asthenia may be related to glottic insufficiency, age (usually present in the elderly) and to the etiology of vocal fold paralysis.^{80,81} Therefore, in this study, asthenia associated with the ML group may be explained because the participants with ML were older (57.59 ± 6.49), which may favor laryngeal alterations, such as the presence of vocal crevices or reduction of vocal fold mucosa, which can lead to glottic insufficiency.³⁰ However, some authors report that clinical manifestation of muscular hypofunction, would not yet be installed in the laryngeal mechanism of voice production,⁸² and also, that uncontrolled pulmonary air may favor the decrease of pneumophonic efficiency⁸³ which may cause voice fatigue,^{84,85} generating asthenia which would involve everybody from the age of 60 years.

On the other hand, vocal instability brings a shivering characteristic to vocal tract structures,^{70,86} which may be related to the decline in muscle strength of the larynx and aerodynamics of the pulmonary current,⁸³ usually linked to the voice of older people,⁸⁷ not to mention the occurrence due to greater adduction of the vocal folds. It is possible that the vocal deviation presented by this group is related to their vocal habits or even due to greater laryngeal constriction. This constriction would reduce transglottic flow, which may lead to pneumophono-articulatory incoordination.⁸⁸ Furthermore, it was verified that 13.63% of these participants presented incomplete vocal coaptation, which may have been compensated by laryngeal hypercontraction (Figure 1).

Concerning the vocal acoustic characteristics, the measure of the variation of the fundamental frequency (vF0), was higher for the participants with ML (Table 4), indicating association of this group, with statistically significant results. These high values may be due to the anatomical characteristics of male larynx, with greater extent and volume of the vocal folds, which favors phonatory instability, once these measures are related to instability of the cycle-to-cycle signal or, in short term and to the control over the

phonatory system.^{44,90} In addition, it was observed that this parameter may be related to hoarseness and voice roughness,⁹¹ which were also characteristics presented by the group with ML, without statistical significance, although with a high percentage and rough voice yet with moderate degree. However, it should be pointed out that these vocal deviations can also be attributed to the age of the participants.³⁰

Furthermore, the frequency measures (STD, F_{hi}) and the number of subharmonic segments (NSH) were also associated with the ML group, with values higher than the CL group and statistically significant. Similar to vF₀, the high result of these measures may also be related to the discrete instability in frequency maintenance.³⁰ This can occur due to resonance alteration, caused by the imbalance in the air direction due to nasal changes (for example, nasal obstruction). Such changes may trigger oral breathing, which will lead to changes in muscle function, leading to compensatory effort in the musculature of the larynx, which would trigger emission instability.⁷³

Three quarters of patients with ML presented higher values of the number of unvoiced segments (NUV) than the CL group, with statistically significant values (Table 4). This measure is characterized by disrupted periodicity of the sound wave, reflected as noise or irregular emission. There are studies that show that higher values of these measures are associated with males.⁹² However, this was not confirmed in this research, once only three quarters of ML participants presented high values and that the CL group was also composed by males. Moreover, people who go through the aging process may exhibit instability of the voice quality and increase of voice breaks and or frequency changes, which would justify the increase in the ML group.³⁰

The patients with CL presented statistically significant association with the noise measure SPI, indicating higher presence of noise to phonation than the ML group. These high values suggest inadequate closure of the vocal folds,^{30,42} or can occur as consequence of bass voices (as in males) and aerial turbulences due to nonlinear phonation.⁹³ To avoid voice disorders, voice exercises can be used, which usually cause increase of the muscle tissue temperature and of the blood flow, reducing the impairment of the muscular work.^{94,95} In this context semioccluded vocal tract exercises can be used. They are performed by partial occlusion of the anterior portion of the vocal tract, which becomes constrict or lengthened, promoting retroflex resonance toward the vocal folds. Some variations of these exercises are described in recent researches, such as nasal sounds^{17,23} used in this work.

After speech therapy intervention, there was improvement of the acoustic parameters in both groups. However, there was statistically significant reduction of the measures of frequency perturbation (vF₀, PPQ, sPPQ) for the CL group (Table 4). As the technique of nasal sounds is an exercise that helps to shift the resonant focus from hypo to hyper, it reduces the tension of the larynx and pharynx,

acting as a springboard of voice projection to the space, which will reduce low resonance and increase the oral component of nasal resonance, producing richer harmonics series and bringing more stability to emission.^{23,96,97}

In addition, there was a statistically significant reduction of the measures of subharmonic segments (DSH, NSH) for the participants with ML (Table 4). The measures of the subharmonic components allow measuring the presence of these low intensity components located between the harmonics. Their decrease after the vocal technique shows reduction of the noise to emission. In the same way, the decrease of the parameters of unvoiced segments, which represent the interruption of the sound wave, indicates reduction of the irregularities during vocalization.⁴⁴ Moreover, there was statistically significant improvement of the frequency measure STD, suggesting greater emission stability (Table 4).

In addition, when only the group with ML was analyzed, before and after the speech therapy intervention (Table 5), improvement of the noise parameters (VTI, NHR, SPI) was observed, indicating regularization in the vibration of the laryngeal structures,^{42,89} which may have occurred due to the benefit brought to vocal folds coaptation, as well as by the better channeling of the aerial flow to the resonance cavities, suggesting improved glottic closure during phonation.⁴² In the same way, the decrease in the parameters of unvoiced segments, which represent the interruption of the sound wave, indicates reduction of the irregularities during vocalization.⁴⁴

Concerning the vocal characteristic of roughness (Table 3), it can be observed that after applying the speech therapy technique, participants of both groups presented reduction of this vocal type, although the reduction of the CL group was greater than that of the ML group, with statistically significant results. Roughness usually occurs in a noisy and unpleasant way, poor in harmonics and rich in noise,⁹⁸ emerging as a consequence of the rigidity of the covering of the vocal folds and or caused by muscle rigidity due to tension increase,⁹⁹ bringing vibration irregularity to emission.⁹⁹ Thus, the reduction of this vocal characteristic may have occurred due to the smoothing of the emission, the reduction of the laryngeal hypertonicity and the improvement of the vibration of the vocal folds, favored by the nasal sounds technique.^{23,96,97}

Finally, we have to point out that this study has some limitations such as the possible interference of presbylarynx on ML group vocal disorders and that the study was conducted only with males. Further studies should use a younger public, and, if possible include the female population, even though ML is more common among males.

CONCLUSION

The purpose of this research was to improve the understanding of the relationship between Leishmaniasis and voice, especially the Mucosal Leishmaniasis. We verified that even without the presence of sequelae of laryngeal

lesions, people with ML may present voice disorders caused by sequelae of nasal lesions or in other sites, which interfere in the correct vocal emission. We highlight asthenia and the most alterations of the acoustic measures (f_{hi}, STD, vF₀, SPI, NSH, DSH) associated with the ML group, which presented the greatest vocal deviations when compared to the CL group. This may have occurred due to the impact of the disease on laryngeal structures, as well as due to voice disorders caused by age and gender.

The CL group also presented vocal changes, marked by vocal instability along with noise measurements (SPI). However, the changes did not present great severity when compared to the ML group. These alterations can be attributed to the incorrect vocal habits of the laryngeal hypercontraction performed in an attempt to compensate for incomplete vocal coaptation.

Despite the disfiguring lesions of ML, which lead to greater vocal impairment of individuals with this disease, we proved that vocal intervention, even over a short period of time, considerably benefits the voice of these patients, improving their quality of life and voice. The results of this study can stimulate the creation of measures of vocal promotion and prevention and better speech therapy strategies. We also verified voice disorders in the CL group, which was not expected, because it was a group characterized by lesions in regions that are not related to the vocal apparatus. However, similar to the ML group, the speech therapy brought significant improvement of the acoustic and auditory perceptual voice parameters.

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