

Oropharyngeal dysphagia in patients with chronic Chagas disease: phonoaudiological, videofluoroscopic, and manometric evaluations

Danielly Moreira Gonçalves Cabral¹, Luiz João Abrahão Júnior², Charles Henrique Dias Marques³, Basílio de Bragança Pereira⁴, Roberto Coury Pedrosa⁵

ABSTRACT

Objective: To determine the frequency of episodes of laryngeal penetration and aspiration in chronic Chagas patients and to evaluate the interpreted relationship not only with the patterns displayed in videofluoroscopy and manometry, but also with clinical screening and structural and functional phonoaudiological evaluation.

Method: Clinical and phonoaudiological (structural and functional) evaluation, fluoroscopy, and computed manometry were performed on all patients. **Results:** Twenty-two patients were included in the study, fifteen females and seven males. Age ranged between 37 and 70 years, mean 55.9 ± 10.2 years. Among the complaints in clinical screening, 18.2% were choking, 13.6% reported throat-clearing, 40.9% heartburn, 22.7% regurgitation, and 36.4% had the sensation of food stuck in the throat. Only 18.2% of patients had adequate dentition. Functional assessment of swallowing detected only 31.8% capable of functional swallowing. In videofluoroscopy, lingering residues were found in the pharynx in 18.2% of cases, followed by multiple swallows 95.4% and 100% posterior escape. There were 4 cases of grade 2 laryngeal penetration (dysphagia) and, in 82% of cases, the findings were similar for the non-occurrence of laryngeal penetration in the videofluoroscopy and in the functional assessment of swallowing. The apertures of the upper esophageal sphincter indicated a relationship with the volume of swallowed bolus. In the manometry, 42.1% of changes were found in the body of the esophagus and 5.3% in the pharynx. **Conclusion:** Laryngeal penetration was prevalent in 18.2% of cases with an important interpretation of the functional relationship between clinical assessment and videofluoroscopic findings regarding the absence of laryngeal penetration, with similar results in 82% of cases.

Keywords: Chagas Disease, Esophageal Motility Disorders, Deglutition Disorders, Manometry, Fluoroscopy

¹ Speech therapist at the Hospital Universitário Clementino Fraga Filho.

² Associate professor at the Universidade Federal do Rio de Janeiro - UFRJ.

³ Assistant professor, Phonoaudiology Course, Universidade Federal do Rio de Janeiro - UFRJ.

⁴ Full professor of Biostatistics and Applied Statistics at the Universidade Federal do Rio de Janeiro - UFRJ.

⁵ Tenured professor, Post-graduation at the Edson Saad Cardiology Institute, Universidade Federal do Rio de Janeiro - UFRJ.

Mailing address:
Roberto Coury Pedrosa
E-mail: coury@hucff.ufrj.br

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INTRODUCTION

Chagas disease or American trypanosomiasis was discovered and described by the physician and epidemiologist Carlos Chagas in 1909. It is a transmissible infection caused by the *T. Cruzi* parasite that can damage the heart as well as the digestive system organs.^{1,2}

The impairment of the digestive tract is manifested mainly in the colon and esophagus, stemming from damages to the ganglion cells in the myenteric plexus and also in the muscle coating of the organ. Its physiopathological base are the changes in motility, especially in the esophagus and colon, with the consequent dilation in the caliber of the final portions of the large intestine, known as megacolon, or of the esophagus, known as megaesophagus.³

Much is described in the literature about dysphagia in the Chagas disease. However, the main comment on the Chagas patient is the esophageal phase that, in advanced cases, can be characterized by the absence of relaxation in the lower esophageal sphincter (LES) and the appearance of simultaneous contractions in the body of the esophagus, which means a loss of peristaltic contractions, generating difficulty in the emptying and the consequent dilation of this organ, creating a megaesophagus. In this phase, many patients refer to the dysphagia as a sensation of food stuck in the retrosternal region, and also as food stuck in the region of pharyngeal recess.⁴

Although esophageal dysphagia is the most investigated alteration in Chagas disease, there are alterations found in the dynamics of oropharyngeal deglutition of Chagas patients that seem to be a result of a compensation mechanism to the alterations in esophageal motility observed in the disease.⁵

The physiological process of deglutition requires the coordinated functioning of the oral cavity and of phono-articulatory organs, pharynx and esophagus, and if one of these components starts to function weakly, it is expected that the others will also be affected.⁶

OBJECTIVE

The present study proposed to determine the frequency of laryngeal penetration and aspiration in a group of patients carrying the Chagas disease, as well as to evaluate the interpreted relationship of the laryngeal penetration and aspiration, not only with the patterns shown by the videofluoroscopy (VFS)

and by the esophageal manometry, but also with a clinical triage and the structural and functional phonoaudiological evaluation.

METHOD

This is an observational, cross-sectional study of a series of non-consecutive cases, made in a tertiary center, in the period between November of 2012 and April of 2014.

The patients were selected in the Chagasic Cardiopathy Clinic of the Cardiology Department of the *Hospital Universitário Clementino Fraga Filho* (HUCFF). The study was approved by the HUCFF-UFRJ Ethics in Research Commission, decision CEP No. 85951, under research protocol N^o 142.100, meeting the national and international directives for research with human beings (Resolution N^o 196/96 of the National Health Council), that regulates experiments involving people.

In this study 7 males and 15 females were included, all aged between 37 and 70 years, with a mean of 56 and standard deviation of ± 9.7 , in active, regular follow-up and with etiological diagnosis of Chagas disease confirmed by two different serological methods. They were asymptomatic and had been living away from the endemic zone for more than 20 years, with their ventricular function preserved or minimally compromised (FEVE > 50%) according to magnetic resonance imaging, in addition to a resting 12-lead electrocardiogram. They then signed the free and informed consent forms.

Patients were not included in the study if they presented with any of the following characteristics: ongoing abusive use of alcohol or illicit drugs (based on DSM IV); evidence of acute systemic infection; chronic obstructive pulmonary disease (COPD) with continuous use of steroids or bronchodilators; pregnancy; patients submitted to head and neck surgeries; presence of other morbidities as observed in their medical records, such as COPD, systemic arterial hypertension, chronic renal insufficiency, orovalvar disease, and neoplasias in any location or of any nature; previous digestive surgeries; presence of neurological disorders or medications that could interfere with the physiology of deglutition.

Once selected, they were submitted to clinical triage, structural and functional phonoaudiological evaluation, videofluoroscopy, and esophageal manometry. The protocols used were based on questionnaires already in the literature and adapted in accordance with the profile of the population studied.^{7,8}

For the clinical triage, a questionnaire was applied to detect the existence of any sign or symptom specific to esophageal dysphagia, which in the case of the present study would be an important datum during the execution and analysis of the referred analyses.

Clinical triage

1. Do you choke frequently?
() no () yes
2. Do you cough? () no () yes If (yes) - is it a night cough, sporadic, or is it related to feeding?
3. Do you have to clear your throat?
() no () yes If (yes) - is it sporadic or is it always related to feeding?
4. Does your voice sound wet during and/or after swallowing?
() no () yes
5. Do you have the sensation of food stuck or of difficulty in swallowing?
() no () yes If (yes) - what type of food: liquid/solid
6. Have you lost weight? () no () yes
7. Do you feel back pains and/or frequent changes in your breathing?
() no () yes
8. Do you need to ingest liquids during meals? () no () yes
9. Do you have the sensation of a full stomach? () no () yes
10. Do you have a bitter taste in your mouth? () no () yes
11. Do you have heartburn frequently?
() no () yes
12. Do you vomit frequently? () no () yes If (yes) - what? (mucus, acid, food)
13. Do you have frequent bad breath? () no () yes
14. Do you usually have a lack of appetite?
() no () yes
15. Do you hypersalivate? () no () yes
16. Do you have frequent hiccups?
() no () yes
17. Do you feel pain when swallowing?
() no () yes

Do you wish to report any other complaint specifically related to eating? () no () yes If (yes) - what would it be?

In the structural analysis, an evaluation of the phono-articulatory organs was made to observe the presence of teeth and their state of preservation, as well as the use of dental prostheses and their adaptation (factors that directly influence the deglutition process; and saliva alterations (subjective evaluation). The symmetry, posture, extra- and intra-oral sensitivity and mobility were also evaluated.⁸

Another important factor described was the evaluation of whether or not a voluntary cough was present to protect the air pathway against the entry of foreign bodies.

However, in the functional evaluation, three types of food consistency were used: fine liquid (water), semiliquid, and solid. The first consistency was offered on a tablespoon with the approximate volume of 10 ml. The semiliquid food was obtained using 15 ml of powdered thickener diluted in 50 ml of water. Finally, a cornstarch cookie was offered as a solid food. These consistencies were also evaluated freely (without determining quantity/volume) to verify the performance of the patient in his normal routine.

The evaluation mechanisms involved not only the clinical observation of the patient, but also the cervical auscultation and the measuring of the peripheral oxygen saturation (SpO₂).

After the clinical triage and obtaining the structural and functional data, the deglutition disorders were classified according to the specific protocol⁹ that classifies deglutition into 7 different levels, the only cases that received a diagnosis of dysphagia were those in which the laryngeal penetration and/or aspiration was evident.

The VFS, with a maximum duration of three minutes and minimum exposure to x-ray, was made after an eight hour fast; the patient was seated, initially at lateral and antero-posterior viewing angles after three deglutitions of barium sulfate at 100%. Each patient was offered, consecutively, the volumes of 3 ml, 5 ml, and 10 ml, first in a glass and then on a tablespoon, as well as, in the semiliquid consistency, obtained mixing 15 ml of powdered thickener diluted into 50 ml of water and 20 ml of barium, offered in a glass with specific measurements. First, it was solicited that a comfortable volume (a comfortable swallow) be ingested and then the rest of the mix (the entire volume). For the solid consistency, we offered a cornstarch cookie dipped into barium.

The equipment used was the image intensifier from Siemens (Siremoibil Compact L, Siemens, Erlanger, Germany). The images were recorded onto a DVD (Phillips do Brasil, São Paulo, Brazil) and then captured on a computer in AVI format to be reviewed in graphic software (Virtualdub) frame by frame.

The abnormalities investigated were the constitution of the dental arch, the presence of intra-oral escape and oral organization, lingual ejection, the presence of palatal escape,

penetration or aspiration through the air pathway, the presence of post-deglutition residue, upper esophageal pharynx-sphincter incoordination, abnormalities in the anteriorization of the larynx, and the presence of pharyngeal bar.

The intraoral escape was considered when the bolus to be ejected was positioned above and below the tongue, taking on the dipper pattern.¹⁰

However, the premature posterior escape was characterized by the premature escape of the food to the hypopharynx, surpassing the region where the pharyngeal response should occur.¹¹

For the degree of penetration and/or aspiration, a scale specific for penetration and aspiration was used,¹² as observed in Chart 1.

The anteriorization of the larynx (cm) and the aperture of the upper esophageal sphincter were also quantified through an image analysis software (Sigma Scan, San Rafael, CA, USA).

The measurements of the maximum elevation and anteriorization of the hyoid bone were obtained from the markings of specific points at the base of the hyoid bone (the lowest point), as well as anchor points in the most anterior region of the mandible, avoiding conflicts in case the patient moved or changed the position of his head.

However, the aperture of the upper esophageal sphincter (USS) was marked through the anteroposterior aperture of the narrowest portion of the aerodigestive tract, between the C5 and C6 vertebrae, during the maximum distension for the passage of the bolus.

The esophageal manometry was done after a four-hour fast, not concomitant to the VFS, with interruption on the day before, in the case of calcium, nitrates, and prokinetic channel blockers being used. A polyvinyl catheter with 4.5mm diameter and eight channels (four distal openings at the same level, positioned radially, and the next four positioned every 5cm) connected to external pressure

transducers and continuously perfused with distilled water at 0.6 ml/min, by a hydraulic capillary pump with low compliance (Alacer Biomédica, São Paulo, Brazil). The intraluminal pressures were recorded by a polygraph (Alacer Biomédica, São Paulo, Brazil) and the digital information transferred to a computer.

The esophageal pathology was characterized by the presence of peristaltic contractions with multiple peaks, segmental aperistalsis of the distal esophagus with relaxation of the normal LES, total aperistalsis with normal relaxation of the LES, incomplete and isolated relaxations of the LES, low amplitude contractions, simultaneous contractions, failures in peristaltic conduction, and prolonged peristaltic contractions. In the UES, the means of the pharynx contractions from the six deglutitions were evaluated, as well as the mean for the lowest point of the UES relaxation, the time of UES aperture, and the synchrony between the pharynx contractions and the UES aperture.

Tables were prepared for the descriptive analysis of the data. The categorical data were expressed in frequency (n) and percentages (%). The numerical data were expressed as the mean \pm standard deviation.

The interpreted relationship regarding the laryngeal penetration and aspiration between the videofluoroscopy and the functional evaluation of the deglutition was verified by the inter-rater agreement.

To analyze the values of the hyoid bone anteriorization and elevation in the objective deglutition examination, the data were analyzed in a statistical package (SPSS v.13), using the *t*-test. The *p* value of 0.05 was adopted for statistical significance.

RESULTS

The study had 22 patients, according to the inclusion criteria already specified. They were submitted to clinical triage, structural

Chart 1. Rosenbeck Scale of Penetration and Aspiration¹²

Category	Score	Description
Penetration	1	Contrast medium does not enter in the air pathway.
	2	Contrast medium enters up to above the vocal chords, with no residue.
	3	Contrast medium remains above the vocal chords, visible residue.
	4	Contrast medium reaches the vocal chords, with no residue.
	5	Contrast medium reaches the vocal chords, visible residue.
Aspiration	6	Contrast medium passes the glottic level, but there is no residue in the subglottic level.
	7	Contrast medium passes the glottic level, with residue in the level, despite the patient's response.
	8	Contrast medium passes the glottis with residue in the subglottis, but with no patient response.

and functional phonoaudiological evaluation, as well as the objective videofluoroscopy exam ($n = 22$). In the esophageal manometry three patients were lost, totaling 19 patients with the complete evaluation process concluded.

The most relevant results found during the phonoaudiological evaluation (clinical triage, structural, and functional) among the 22 participants of the study are shown in Tables 1, 2, and 3, respectively.

Attention should be given to the bad dental state in 82% of the population studied, which could interfere directly with the oral organization process.

In the functional evaluation, seven patients (31.8%) received the functional deglutition diagnosis (Table 3). This happened because during the diet evaluation, they presented the complaints of food stuck in the throat (13.6%), presence of multiple deglutitions (13.6%), cough after deglutition (18.2%), and throat clearing (9.1%), without any occurrence of laryngeal penetration being evident through the cervical auscultation.

In the videofluoroscopy, the permanence of residues in the pharynx, after deglutition, was observed only in four patients (18.2%) and through isolated occurrences such as: liquid consistency (glass and spoon), one single case in the pasty consistency and another in the solid consistency. The other occurrences can be seen in Table 4.

Episodes of laryngeal penetration occurred in four patients (18.2%), all with the volume of 10 ml (barium/glass and spoon utensils) and they received grade 2. Thus, episodes of laryngeal penetration and/or aspiration were not observed in 18 patients, that is, 82%. According to the characteristics demonstrated in the VFS, 18.2% of the cases were diagnosed as discreet oropharyngeal dysphagia.

As for the measurements obtained from the VFS images, the means for the elevation and anteriorization of the hyoid bone and UES aperture were found. No discrepancies or direct relationships were observed between the means for the elevation and anteriorization of the hyoid bone with the aliquots and

consistencies offered. However, for the UES aperture, a relationship was observed with the volume offered for ingestion, showing a tendency to increase the aperture as the volume of bolus is increased (Table 5).

In the relationship between the VFS and the functional phonoaudiological evaluation, it was observed that, of the 22 patients, the findings were similar in 82% of the cases for the non-occurrence of laryngeal penetration. In other words, in only four cases was there any discrepancy between the objective examination that revealed grade 2 for laryngeal penetration and the functional evaluation of deglutition, in which it was not observed.

As for the esophageal manometry, we observed that in 42.1% (8/19) of the cases, there were alterations in the body of the esophagus, such as hypomotility (seven occurrences) or a hypotense LES (three occurrences). Only in one single case, that is, 5.3%, did this alteration occur in the pharynx region, with weak contractions, and it was not found in the videofluoroscopy.

In the relationship between the clinical triage and the esophageal manometry, it became clear that the patients with clinical complaints compatible with the low involvement of the esophagus such as heartburn, regurgitation, sensation of food stuck, presence of multiple deglutitions, coughing and throat clearing after eating, had an altered manometry (hypomotility, hypotense LES, and weak contractions in the pharynx region), which was in 42.1% of the cases (8/19).

However, in the observation between the functional evaluation of deglutition and the esophageal manometry, two patients presenting coughing after deglutition, sensation of food stuck, and throat clearing, also presented manometric findings, both with hypomotility in the body of the esophagus.

Between the videofluoroscopy and the esophageal manometry, it was observed that of the four individuals who presented laryngeal penetration, only two showed corresponding findings with manometry, although with alterations only in the body of the esophagus.

DISCUSSION

As mentioned before, Chagas-related dysphagia involves the esophagus. Among the patients infected with the protozoan, 7 to 10% will develop the esophagus disease shown by the symptoms of dysphagia, regurgitation, thoracic pain, and abnormal motility with simultaneous contractions in the body of the

Table 1. Clinical triage of 22 patients

Complaints	Number	Percentage
Heartburn	09	40.9%
Liquids with meals	05	22.7%
Sensation of food stuck for liquids and solids	05	22.7%
Regurgitation	05	19%
Choking	04	18.2%
Throat clearing after eating	03	13.6%
Coughing while eating	03	13.6%
Sensation of food stuck only for solids	03	13.6%
Wet voice	0	0%
Respiratory alterations	0	0%
Hypersalivation	0	0%

Table 2. Structural phonoaudiological evaluation of the 22 patients

Items Researched	Number	Percentage
Spontaneous deglutitions of saliva	22	100%
Appropriate tongue mobility	22	100%
Appropriate soft palate mobility	22	100%
Occlusion of lips at rest	22	100%
Preserved intraoral sensibility	22	100%
Strong coughing	22	100%
Normal perception of saliva	19	86.4%
Normal vocal quality	14	63.6%
Hoarse voice quality	8	36.4%
Teeth in good condition	4	18.2%
Excessive, thick, or fluid saliva	3	13.6%
Alterations in the oral cavity	3	13.6%

Table 3. Phonoaudiological diagnosis according to functional evaluation of the 22 patients

Phonoaudiological Diagnosis	Percentage
Normal deglutition	68.2%
Functional deglutition	31.8%
Discreet dysphagia	0%
Discreet/moderate dysphagia	0%
Moderate dysphagia	0%
Moderate/severe dysphagia	0%
Severe dysphagia	0%

Table 4. Videofluoroscopic occurrences

Occurrences	Consistency	Percentage
Premature posterior escape	Solid	100%
Premature posterior escape	Pasty	59.1%
Multiple deglutitions (about 3)	Solid	90.9
Multiple deglutitions (about 3)	Liquid (10 ml of barium/glass)	45.4%
Intraoral escape	Liquid (10 ml of barium/glass)	31.8%
Intraoral escape	Pasty	31.8%

Table 5. Means for anteriorization, elevation, and aperture of the UES (n = 22)

	Mean Total (cm)		
	Anteriorization	Elevation	Aperture
3 ml glass	1.02	0.74	0.6
5 ml glass	1.09	0.68	0.65
10 ml glass	0.92	0.94	0.89
3 ml spoon	1.04	0.61	0.65
5 ml spoon	1.03	0.82	0.79
10 ml spoon	1	0.81	0.84
Comfortable swallow	1.05	1.06	1.03
Total Volume	0.83	0.59	0.88
Cookie	1.14	1.06	0.86

esophagus, as well as the absence or partial relaxation of the LES.^{13,14} These symptoms can start weeks or even years after the infection, in a slow and progressive manner for solid and liquid foods.¹⁵

Among all the clinical manifestations of esophageal disorders, this work focuses on the oropharyngeal disorder, a complication with the potential for morbidity and mortality, especially in relation to the pulmonary function. Although the biological evidence indicates a worse prognosis associated with chemical pneumonia as a result of high dysphagia, the real meaning and relevance of this phenomenon still needs more definitive studies.

In the present study, the absence of laryngeal penetration in the functional

phonoaudiological evaluation could not be compared with previous studies, for such occurrence was not investigated among the works involving Chagas-related clinical phonoaudiological evaluation.^{13,16}

Nevertheless, in the VFS, intraoral escape was found in 54.5% of the cases, which may be justified in our study by the bad state of the patients' teeth (82%), causing a deficient oral organization.

The lingering of pharyngeal residues, found in 18.2% of the patients, is similar to the findings in previous studies,^{14,17,18,19} which showed a longer pharynx transit and depuration time in patients with the Chagas disease.

The occurrence of multiple deglutitions in the present study is also described in recent

works,^{13,16} which found a greater number of deglutitions and a longer interval between them. However, the occurrence of premature posterior escape in 100% of the cases for solid consistency, although significant, it not mirrored by results in the literature involving the Chagas disease, as a form of comparison. Other than giving a better clinical characterization of the Chagas disease patients, these works give no information on the diagnostic capacity evaluated by inter- and intra-rater agreement.

The studies found on the videofluoroscopic evaluation on the Chagas disease discard the occurrence of laryngeal penetration and/or aspiration for the population studied.^{14,19} However, episodes of laryngeal penetration (grade 2) were observed in 18.2% of the cases in the present study. Before establishing that the events above are rare in patients with the Chagas disease, it would be necessary to evaluate the definitions used for "penetration" and "aspiration" in the referred studies, determining whether there is a uniformity for the diagnostic criteria. In the same way, it is noteworthy that the evidence of laryngeal penetration (grade 2) can also be found in normal individuals, thus showing the need for correlation with the treatment.²⁰ Therefore, such penetration in healthy individuals should be characterized by the absence of residues after deglutition, becoming a problem only for those penetrations where the subject cannot expel the food from the airway.²¹

Another relevant finding was the interpreted relationship between the VFS and the functional clinical evaluation of the deglutition for the absence of laryngeal penetration, with similar findings in 82% of the cases, a value even greater than the one found in a previous study that also compared the clinical evaluation (cervical auscultation) and the VFS, having an inter-rater agreement of 76%.²²

The measurements made based on the VFS are in agreement with previous findings,²³ since a direct relationship between the UES aperture and the volume to be swallowed was also found. This behavior is no different from a normal population, according to the same author.

As for the esophageal manometry, previous studies with asymptomatic patients, only with positive serology for the Chagas disease, showed the occurrence of simultaneous contractions, failure in the relaxation of the LES, and low amplitude contractions.²⁴ Later, the occurrence was also observed of a

greater interval between the beginning of the pharyngeal contractions and the beginning of the proximal esophageal contractions.¹⁶

Finally, the alterations found in the dynamics of oropharyngeal deglutition, verifying the diagnosis of discreet oropharyngeal dysphagia in 18.2% of the cases, could become a response of a compensation mechanism to the possible alterations of esophageal motility, which are common with the disease and already described and observed in the literature.⁵ However, for the group studied, the esophageal manometry showed alterations (body of the esophagus) in only eight patients (42.1%), with correlation between the manometric and videofluoroscopic findings in only two cases.

Moreover, it is important to note the bad state of the teeth observed in most of the patients in this sample—a factor that can also compromise the oral phase of deglutition, creating an inefficient ejection and increasing the risk of aspiration, showing that the oral organization affects not only the quality of oral ejection, but also the effective dynamic of the pharyngeal phase.¹⁰

The limitations found in this study, such as the small number of individuals in the sample and the absence of a control group, did not alter or damage the results of the work, since it is a case series study, which offers the possibility of continuity through supplementary studies.

CONCLUSION

Laryngeal penetration was prevalent in 19.2% of the cases with an important interpreted relationship between the functional phonocardiological evaluation and the videofluoroscopic findings for the absence of laryngeal penetration with, similar results in 82% of the cases.

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