CoDAS
ISSN 2317-1782 (Online version)

Original Article
Artigo Original

Correlation between bulbar functionality and laryngeal penetration and/or laryngotracheal aspiration on motor neuron disease

Correlação entre funcionalidade bulbar e penetração e/ou aspiração laringotraqueal na doença do neurônio motor

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Keywords

Motor Neuron Disease
Deglutition Disorders
Videofluoroscopy

ABSTRACT

Objective: Describe and correlate bulbar functionality with laryngeal penetration and/or laryngotracheal aspiration for different food consistencies in Motor Neuron Disease (MND). **Methods:** Study participants were 18 individuals diagnosed with MND regardless of the type and time of onset of disease. The Amyotrophic Lateral Sclerosis Functional Rating Scale - Revised/BR (ALSFRS-R/BR) was applied, and only the bulbar parameter, which includes speech, salivation and swallowing, was analyzed, with scores raging from 0 (disability) to 12 (normal functionality). Swallowing videofluoroscopy was performed using the Penetration-Aspiration Scale (PAS) described by Rosenbek et al. (1996). The Pearson correlation test was used for data analysis. **Results:** According to food consistency, the PAS level ranged from 1 to 5 for puree consistency, 1 to 4 for thickened liquid, and 1 to 3 for liquid, and no laryngotracheal aspiration was observed. Negative correlation between bulbar functionality and laryngeal penetration was observed for all food consistencies (pasty: r=-0.487, p=0.041; thickened liquid: r=-0.442, p=0.076; liquid r=0.460, p=0.073), but statistically significant difference was found only for the puree consistency, that is, individuals with poor bulbar functionality presented higher levels of laryngeal penetration. **Conclusion:** Negative correlation was observed between bulbar functionality and laryngeal penetration in MND. The bulbar parameters of the ALSFRS-R/BR are significant for predicting risk of laryngotracheal aspiration for pasty consistency in MND.

Descritores

Doença do Neurônio Motor
Transtornos de Deglutição
Videofluoroscopia

RESUMO

Objetivo: Descrever e correlacionar a funcionalidade bulbar com penetração e aspiração laringotraqueal em distintas consistências de alimento na Doença do Neurônio Motor (DNM). **Método:** Participaram do estudo 18 indivíduos diagnosticados com DNM, independentemente do tipo e tempo da doença. Foi aplicada a escala *Amyotrophic Lateral Sclerosis Functional Rating Scale–Revised/BR* (ALSFRS-R/BR), sendo analisado apenas o parâmetro bulbar que compreende fala, salivação e deglutição, com pontuação de 0 (incapacidade) a 12 (funcionalidade normal). Realizou-se videofluoroscopia da deglutição com aplicação da *Penetration-Aspiration Scale (PAS)* descrita por Rosenbek et al. (1996). Realizado Teste de Correlação de Pearson. **Resultados:** Considerando a consistência do alimento, o nível da PAS variou de 1 a 5 na consistência pastosa, de 1 a 4 na líquida espessada e de 1 a 3 na líquida rala, e não houve aspiração laringotraqueal. Para todas as consistências de alimentos, houve correlação negativa entre funcionalidade bulbar e penetração laríngea (pastoso: r=-0,487, p=0,041; líquido espessado: r=-0,442, p=0,076; líquido ralo: r=-0,460, p=0,073), porém somente na consistência pastosa houve diferença estatística significante, ou seja, indivíduos com baixa funcionalidade bulbar apresentaram maior nível de penetração laríngea. **Conclusão:** Houve correlação negativa entre funcionalidade bulbar e penetração laríngea na DNM. Os parâmetros bulbares da escala ALSFRS-R/BR mostraram-se significantes para predizer risco de penetração laringotraqueal na consistência pastosa na DNM.

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Received: March 20, 2017

Accepted: August 19, 2017

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Financial support: nothing to declare.

Conflict of interests: nothing to declare.

INTRODUCTION

Motor Neuron Disease (MND) characterizes clinical syndromes in which involvement of the lower and/or upper motor neurons is observed, such as Amyotrophic Lateral Sclerosis (ALS), Progressive Muscular Atrophy (PMA), Primary Lateral Sclerosis (PLS), and Progressive Bulbar Palsy (PBP)[1,2].

These syndromes can cause dysphagia, and they affect up to 85% of individuals with ALS[3]. Oropharyngeal dysphagia in this population is associated with progressive degeneration of the corticobulbar pathways to IX, X, XI and XII cranial nerve motor nuclei, causing secondary alterations in pharyngolaryngeal contraction and atrophy of the vagus and glossopharyngeal nerves, leading to difficulty in the palatine veil movement and decrease in laryngeal elevation, as they are responsible for the motor and sensory innervation of the larynx and pharynx[4]. In addition, oropharyngeal dysphagia in this population is marked by changes in the oral phase of swallowing, which is justified by weakness, fasciculation, and tongue atrophy, thus determining oral incoordination with impairment to feeding[4].

Oropharyngeal dysphagia can occur regardless of the type of disease onset, either bulbar or spinal, with presence of laryngeal penetration and laryngotracheal aspiration in patients with MND[3-6].

There has been frequent search for MND progression indicators[7]. Currently, there are scales that assist with monitoring the symptoms of these patients and providing specific parameters for each motor function.

Considering that, during evolution of MND, bulbar functions such as speech, salivation and swallowing, as well as motor function in general are impaired due to cranial nerve involvement, individuals with compromised bulbar function may present more severe dysphagia with presence of laryngeal penetration and laryngotracheal aspiration as the disease progresses. Identifying the moment at which individuals with MND present greater risk of laryngeal penetration and laryngotracheal aspiration can assist with the definition of therapeutic behaviors, as well as with the management of symptoms, avoiding nutritional and pulmonary complications and improving quality of life.

In view of the foregoing, the present study aimed to describe and correlate bulbar functionality with laryngeal penetration and laryngotracheal aspiration in patients with MND.

METHODS

This study is part of the project "Association of Global Motor Performance with Oropharyngeal Dysphagia in Amyotrophic Lateral Sclerosis", which was approved by the Research Ethics Committee of the Faculdade de Medicina de São José do Rio Preto (CAAE 53663516.0.3001.5415).

After receiving the necessary guidance and clarifications on the procedures to be conducted, all participants and/or caregivers signed an Informed Consent Form (ICF) prior to study commencement.

Study participants were 18 individuals (14 men and four women) aged 31-87 years (mean=55.66) diagnosed with Motor Neuron Disease (MND) by means of clinical examinations, regardless of the type and time of onset of disease. Of the individuals included in the study, 83.33% (N=15) were diagnosed with Amyotrophic Lateral Sclerosis (ALS), 11.11% (N=2) with Primary Lateral Sclerosis (PLS), and 5.55% (N=1) with Progressive Muscular Atrophy (PMA). Time of disease onset varied from six months to 10 years.

The Amyotrophic Lateral Sclerosis Functional Rating Scale – Revised/BR (ALSFRS-R/BR), translated and validated for Brazilian Portuguese[8], was applied to all participants and/or caregivers, in a private room, by a speech-language pathologist who read the options to the respondents, took notes of their answers, and posteriorly analyzed the results, ensuring the assessment standardization.

The ALSFRS-R/BR evaluates the bulbar function (speech, salivation, and swallowing), fine motor function (handwriting, cutting food and handling utensils, and dressing and hygiene), gross motor function (turning in bed and adjusting bed clothes, walking, and climbing stairs), and respiratory function (orthopnea and respiratory insufficiency) by scores ranging from zero to four for each item and a total score from zero (disability) up to a maximum score of 48 points (normal functionality). However, in this study, only items referring to the bulbar function (speech, salivation, and swallowing) were used, with scores ranging from zero (disability) to 12 (normal functionality).

Objective evaluation of swallowing was conducted by videofluoroscopy in the Radiology Department of the Hospital. The examinations were performed by a radiologist, a radiology technician, and a speech-language pathologist with experience in this procedure. The individuals remained seated and the images were taken in the lateral position with limits marked as follows: upper and lower, from the oral cavity to the esophagus; anterior, by the lips; posterior, by the wall of the pharynx; superior, by the nasopharynx; and inferior, by the cervical esophagus[9]. A Flexavision-HB (Shimadzu Corporation) remote-controlled X-ray device coupled to a video converter was utilized.

The examination protocol began with the standardization of preparation of the consistencies. For puree consistency: 40 ml of water + 15 ml of barium sulfate ($BaSO_4$) + 1 measure of food thickener; for thickened liquid consistency: 20 ml of water + 20 ml of barium sulfate ($BaSO_4$). All consistencies were offered in a volume of 5 ml each, in a plastic spoon, with three 5 ml offers: first in puree consistency, followed by thickened liquid and thinned liquid; the supply was interrupted in the presence of laryngotracheal aspiration. In this study, the videofluoroscopy protocol used only the Penetration-Aspiration Scale (PAS)[10], which presents eight levels ranging from no laryngeal penetration (level 1) to

presence of unresponsive aspiration from the patient, i.e., silent aspiration (level 8).

The correlation study between the bulbar parameters of the ALSFRS-R/BR and the levels of the PAS were performed individually for each food consistency, and the results were submitted to statistical analysis using the Pearson's correlation test, adopting a significance level of 95% ($p \leq 0.05$).

RESULTS

Table 1 shows the classification of individuals with Motor Neuron Disease (MND) according to the Penetration-Aspiration Scale (PAS) levels for each food consistency. No laryngotracheal aspiration was observed in any of the individuals in this sample.

ALSFRS-R/BR scores for the bulbar functionality are depicted in Figure 1. They range from one to 12, with fewer individuals near poor functionality or disability and more individuals close to normal functionality.

Table 2 presents the correlation between bulbar functionality and laryngeal penetration for all food consistencies. Negative correlation was found for all food consistencies, but significant difference was observed only for the pasty consistency (p=0.041).

Figure 2 shows the individual distribution according to bulbar functionality and presence of laryngeal penetration.

Table 1. Classification by the Penetration-Aspiration Scale (PAS) of individuals with Motor Neuron Disease (MND) according to food consistency

Individual	Puree	Thickened liquid	Liquid
1	5	2	2
2	1	4	_
3	1	1	1
4	1	1	1
5	1	2	2
6	3	3	2
7	1	1	1
8	1	1	1
9	1	1	1
10	1	1	1
11	1	_	1
12	2	2	-
13	1	1	1
14	1	1	1
15	1	1	1
16	2	2	3
17	1	1	3
18	1	3	3

Table 2. Correlation between bulbar functionality and laryngeal penetration according to food consistency

Bulbar functionality	Laryngeal penetration Puree		Laryngeal penetration Thickened liquid		Laryngeal penetration Liquid	
	Absent	Present	Absent	Present	Absent	Present
1		1		1		1
2	1			1		
5	1	1	1	1	1	1
6		1		1		1
7	1		1		1	
8	2		2		2	
9	2	1	2	1	2	1
10	2			1	1	1
11	4		3	1	3	1
12	1		1		1	
Pearson's correlation test	-0.487		-0.442		-0.460	
p-value	0.041*		0.076		0.073	

*Statistically significant

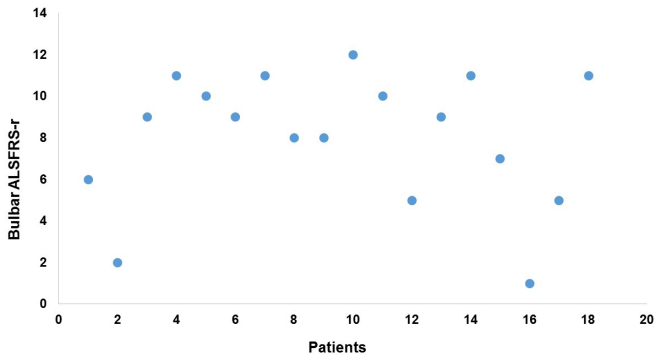

Figure 1. Distribution of individuals with MND in the Amyotrophic Lateral Sclerosis Functional Rating Scale - Revised/BR (ALSFRS-R/BR) for bulbar functionality

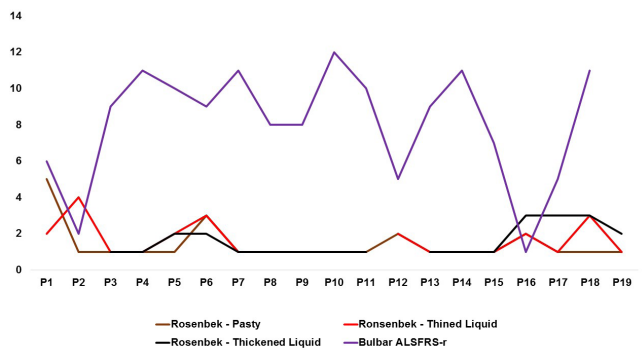

Figure 2. Distribution of individuals with MND in the ALSFRS-R/BR by bulbar functionality and laryngeal penetration according to food consistency

DISCUSSION

Motor Neuron Disease (MND) is strongly characterized by significantly compromising the overall and oropharyngeal motor controls, resulting in multiple damages to the general functional condition, as well as to the safety and efficiency of feeding in this population. Moreover, there are several risk factors to the quality of life of patients with MND, and the presence of oropharyngeal dysphagia is one of these risk markers.

According to the specific scientific literature, in addition to being a frequent comorbidity in MND, dysphagia affects the oral and pharyngeal phases, regardless of food consistency[11]; however, patients with bulbar onset present impaired swallowing in the initial phase of the disease[12].

Presence of oropharyngeal dysphagia in the different underlying diseases, including the MNDs, may contribute to development of pulmonary complications and, therefore, presence of laryngeal penetration and laryngotracheal aspiration has become one of the most relevant parameters to be investigated in the context of clinical and objective assessment of this swallowing disorder. Nevertheless, although there are frequent reports in the literature on the presence of this symptom in MND[4,6], no individuals with laryngotracheal aspiration were found in the sample of this study. However, presence of laryngotracheal aspiration in MND is closely associated with the damage that this disease causes to the oropharyngeal functions and, at a certain moment, with its progression. Sometimes, depending on the type of MND, the oropharyngeal condition is severely compromised since the onset of disease, or it worsens with its progression. Furthermore, a study on MND showed that the presence of laryngotracheal aspiration in this population is related to the onset of bulbar symptoms for more than 24 months[12] and the sample of this study was heterogeneous from the bulbar point of view.

In contrast, different levels of laryngeal penetration were found in the individuals with MND for all food consistencies, evidencing presence of impairment in the oral and pharyngeal phases of swallowing. The different oropharyngeal pathophysiological mechanisms affected by MND begin with the presence of weakness in the tongue musculature, which causes difficulties in formation, containment, and propulsion of the food bolus, affecting pharyngeal response and triggering posterior oral leakage for all food consistencies, especially for liquids[12,13-15]. Another common pathophysiological aspect in MND that contributes to presence of laryngeal penetration and risk of laryngotracheal aspiration is pharyngeal residue, determined by poor oral propulsion and weakness in pharyngeal contraction[16].

Negative correlation was observed between bulbar functionality and presence of laryngeal penetration for pasty consistency, that is, the poorer the bulbar functionality, the higher the level of laryngeal penetration. Other studies that evaluated functionality using the ALSFRS-R/BR or other functional scales have also evidenced this finding[4,17]. This result suggests that the bulbar parameter of the ALSFRS-R/BR can be used as an instrument to suggest risk of laryngeal penetration in MND, corroborating the literature[12].

The bulbar parameters of the ALSFRS-R scale observed in patients with MND can assist with predicting risk of laryngeal penetration, aiding in the early diagnosis of oropharyngeal dysphagia.

CONCLUSION

The levels of laryngeal penetration, as well as bulbar functionality, in Motor Neuron Disease (MND) varied for all food consistencies in the study population. In addition, negative correlation was observed for pasty consistency - the poorer the bulbar functionality in MND, the higher the level of laryngeal penetration.

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Author contributions

BCB and ASG collected, classified, analyzed, and interpreted the data and assisted with the writing of the manuscript; LGL, GST, and MAOMS participated in the writing of the manuscript and critical review of relevant intellectual content; RGS was responsible for the study design, collection, analysis, and interpretation of the data, and writing of the manuscript.