Assessment of Dysphagia in Patients with Multiple Sclerosis

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Original Article

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ABSTRACT

Objectives: This study was conducted to estimate the prevalence of swallowing impairment in Relapsing-remitting multiple sclerosis (RRMS) cases and determine if there is any correlation between disability status and the severity of swallowing impairment to take the results into consideration while drawing the intervention plan.

Patients and Methods: The study included 40 patients diagnosed with RRMS, and they were subjected to history taking, neurological examination, and Fiberoptic Endoscopic Evaluation of Swallowing (FEES), The Yale pharyngeal residue severity rating scale was used.

Results: FEES revealed that 50 % of RRMS patients had no residue, 35% had mild residue and 15% had moderate residue. Additionally, 15% of the patients had choked with penetration, but there was no aspiration or nasal regurgitation. There was no statistically significant difference between the three groups of FEES findings regarding the expanded disability status scale (EDSS). The incidence of swallowing impairment increased with disease duration > 1 year, EDSS > 3, and in patients with more than 2 relapses.

Conclusion: About half of the RRMS patients have pharyngeal residue post-swallow with variable degrees. Disease duration >1 year, a number of relapses >2 relapses, and EDSS >3 are significant risk factors for choking in MS patients. Keywords: Multiple Sclerosis, swallowing, swallowing impairment, fiber optic endoscopic evaluation of swallowing, EDSS.

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INTRODUCTION

The swallowing process consists of four phases: the oral preparation phase, the oral propulsive phase, the pharyngeal phase, and lastly the oesophageal phase[1,2]. Dysphagia is defined as a swallowing difficulty due to weakness, impaired coordination, or obstruction affecting normal swallowing mechanisms[3]. Two terms have been proposed to describe abnormalities in airway protection associated with eating and drinking, “penetration and aspiration”. Penetration describes the condition when the bolus enters the larynx to the level of true vocal folds; whereas aspiration occurs when the bolus moves below the true vocal fold level and enters the trachea. When aspiration occurs before, during, or after swallowing without cough production, it is called “silent aspiration”[4].

Multiple sclerosis (MS) is the most common inflammatory autoimmune disorder of the central nervous system (CNS) affecting mostly young adults[5]. Its incidence shows a worldwide increase, which in turn will lead to an increase in its socioeconomic impact[6]. Although genetic-environmental interaction is believed to play a major role in the pathogenesis of that disorder, the exact mechanisms behind that disease remain opaque[7].

Relapsing-remitting subtype (RRMS) is considered the most common subtype of multiple sclerosis, as not less than 85% of these patients are diagnosed as RRMS. The most common features of PRMS are exacerbation and remission of symptoms (attacks) within days to weeks. In the final stage of each attack, many symptoms fade either completely or partially and remain stable till the following attack. Most cases of RRMS continue as a secondary progressive subtype within 10-20 years. This disease is characterized by localized areas of inflammation, demyelination, and axonal loss along with gliosis in the brain and spinal cord[8], resulting in a wide range of

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manifestations including physical, mental, and sometimes psychiatric symptoms[9]. Patients with MS could report some symptoms of dysphagia. Many factors could lead to the development of such problems in MS patients, including impairment of corticobulbar tracts, cerebellum, brainstem, and/or lower cranial nerves. Dysphagia could be reported in adults with MS who have mild disability levels and its severity increases in patients with moderate or severe impairment[10]. This problem could lead to dreadful health consequences like aspiration, subsequent bronchopneumonia, and mortality, especially in patients with late disease stages[11].

Although swallowing impairment is a life-threatening condition and among the leading cause of death in people with MS, little is known about the exact frequency of swallowing impairment in MS[12]. Thus, we conducted the present study to estimate the prevalence of swallowing impairment in RRMS cases and to determine if there is any correlation between disability status and the severity of swallowing impairment to take the results into consideration while drawing the intervention plan.

SUBJECTS AND METHODS

This cross-sectional study included 40 patients diagnosed as having relapsing-remitting sclerosis (RRMS). The patients were recruited sequentially from the Neurology clinic at Beni-Sue University from February 2019 to October 2019. Before participating in the study, informed oral consent was obtained from cases who accepted to participate in this study after a complete explanation of the steps and complications of every procedure. The study was conducted in accordance with the Declaration of Helsinki. The study was approved by the Ethical Committee, Faculty of Medicine, Beni-sue University, Egypt (Approval number: FMBSUREC/01092020/Ismail).

Inclusion criteria

patients in this study met McDonald's criteria 2017 for diagnosis of RRMS [13]. They were examined during remission states (at least one month following the last relapse). Their age range was between 15 and 45 years.

On the other hand, patients with systemic comorbidities (chronic liver or renal disease, autoimmune disorder, bleeding diathesis, or malignancy), concomitant neurological disorders, and/or dysphagia of other known causes were excluded.

All patients were subjected to selected items of the protocol of assessment of oropharyngeal dysphagia, developed in the Unit of Phoniatrics, Beni-Suef University Hospital. The following protocol was applied:

Neurological Assessment

All patients were subjected to full neurological assessment, done by expert neurologists, including history taking with special emphasis on MS duration, the number of the attacks, and assessment of disability using the expanded disability status scale (EDSS)[14]. The evaluated functional systems were pyramidal, sensory, cerebral, brain stem, visual, cerebellar, bowel and bladder. The score ranges from zero (normal) to 10 (death due to MS). Other neurological assessments included cranial nerves examination especially trigeminal nerve (tingling, numbness, and poor mastication), facial nerve (decreased taste sensation, accumulation of food on one side, and drooling of saliva), glossopharyngeal, vagus, accessory nerves (nasal regurgitation, nasal tone of speech, dysphonia, and dysphagia), and hypoglossal nerve (impairment of tongue movement during eating and speaking).

Elementary Diagnostic Procedures (bedside clinical assessment)

This included history taking and physical examination. Both were important tools in the diagnostic classification of swallowing disorders, swallowing problems, and nutritional status. Other system involvement was also assessed.

Swallowing problems and nutritional status were assessed by asking about: the type of difficulty, difficult food consistency either solid or semisolid or liquid, problems with chewing, difficulty initiating swallowing, nasal regurgitation, drooling, coughing or choking episodes during eating, food sticking in the throat, and voluntary forms of compensation (alteration of dietary characteristics and eating methods).

The development of eating habits was also assessed by asking about cutting food into smaller pieces, chewing more thoroughly, washing down solids with liquids, throat clearing during meals, head-turning or tilting during eating, taking longer to finish a meal, weight loss, self-feeding (no assistance – needs assistance), and if the patient has changed his/her diet because of swallowing difficulty.

Regarding other system involvement, a history of the respiratory system was collected by asking the patient about frequent chest infections, coughing, respiratory distress, aspiration pneumonia, and previous need for tracheostomy.

The previous history of medical disorders, transient ischemic attacks, heart diseases, irradiation, trauma, surgery, and drug intake was collected for all cases.

The oral phase of swallowing was also evaluated; it included observation of patients during eating, their reaction to food, self-feeding skills, and oral movements in chewing.

Clinical Diagnostic Aids

Fiberoptic Endoscopic Evaluation of Swallowing (FEES) was done by expert Phoniatricians, and included a complete examination of the pharyngeal swallowing. It included an assessment of the anatomy and the physiologic function of the selected structures before swallowing followed by a direct assessment of the swallowing function.

Anatomic-physiologic assessment

The technique of Langmore et al. 1988 was applied which started with the passage of the endoscopy through the nose to assess the velopharyngeal port and ends with a complete laryngeal assessment[15].

Swallowing of food and liquids

At this point, measured quantities of liquids, semisolids, and solids were given to the patient to swallow and the findings of the examination included oral stage, residue, together with penetration, and aspiration were collected.

The Yale Pharyngeal Residue Severity Rating Scale is an important tool used to assess the residue site and severity based on FEES, it is a 5-point scale that depends on the residue site (vallecula and pyriform sinus) and size (none, trace, mild, moderate, and severe).

Penetration aspiration scale (PAS) was also used to assess the degree of penetration and/or aspiration that occurred during this study[16].

Sample size calculation

The current sample size was calculated using G. Power version 3.1 software program for windows. It was supposed that the prevalence proportion of swallowing impairment in PRMS patients at a constant proportion (50%), at an effect size (0.29), an alpha error (0.05), a power of the study (97%) with Exact test (difference proportion from a constant), then the sample size was 40 patients with PRMS

Statistical analysis

SPSS v. 25 (Statistical Package for Social Science) for Windows was used for data analysis. The mean and standard deviation (SD) were used to represent quantitative data, whereas numbers and percentages were used to express categorical data. The chi-square test was performed to compare categorical variables between the groups included in the study. One-way ANOVA test was used to examine statistical differences between the subgroups included in the study. In a post hoc analysis, multiple comparisons among groups were subjected to the Tukey test. Quantitative variables were correlated using the Spearman correlation coefficient (r). When the *P-value* is greater than 0.05, it is considered non-significant, and when it is less than or equal to 0.05, it is considered significant.

RESULTS

The mean age of the included cases was 30.5 years. Most of the included cases were females (85%). (Table 1)

Table 1: Demographic data of included patients

Characteristics		Patients (n=40)
Age [Mean (SD)]		30.5±6.7
Sex	Female [N (%)]	34(85%)
	Male [N (%)]	6(15%)

During bedside assessment, all patients were normal, and no abnormalities were detected. Results of FEES findings stated that the included cases were divided into three groups: group without residue (20 cases), group with mild residue in vallecula (14 cases), and group with moderate residue in vallecula (6 cases). Also, only 6 cases had penetration, scored as (2) on PAS. These data were summarized in (Table 2).

Table 2: Findings of FEES in MS patients

			Patients (n=40)
FEES	Residue	None [N (%)]	20 (50%)
		Mild residue in vallecula [N (%)]	14(35%)
		Moderate residue in vallecula [N (%)]	6(15%)
		Penetration [N (%)]	6(15%)
		Aspiration [N (%)]	0
		Choking [N (%)]	6(15%)
		Nasal regurgitation [N (%)]	0

Descriptive of categorical variables (proportion)

The comparison revealed a statistically significant difference between the three groups as regards the patient's age at disease onset. It was significantly older in cases in the moderate group when compared to cases with no or mild residue. However, there was no statistically significant difference between the three groups regarding age, disease duration, or the number of relapses. These data are summarized in (Table 3).

Table 3: Effect of MS characteristics on FEES findings

Characteristics	None (n=20)	Mild residue in vallecula (n=14)	Moderate residue in vallecula (n= 6)	*P-value*
¶ Age [Mean (SD)]	29.8±4.6	30.4±8.8	33.3±8	0.535
¶ Age at disease onset in years [Mean (SD)]	24.9±4.2	25.4±5.2	32.3±8a, b	0.013*
£ Disease duration in years [Mean (SD)]	4.8±4.7	5.2±5.102	1±0	0.156
£ Number of relapses throughout disease duration [Mean (SD)]	3±1.8	3.1±2	1.8±0.7	0.333
£ EDSS [Mean (SD)]	£3.1±1.4	2.8±1.3	2.8±0.6	0.717

¶: One Way ANOVA

£: Kruskal Wallis

A: significant value compared to cases with no residue.

B: Significant value compared to cases with mild residue in the vallecula.

As shown in (Table 4), patients with disease duration >1 year had a significantly higher incidence of choking when compared to patients with disease duration ≤ 1 year ($P = 0.003$).

Table 4: Effect of disease duration on choking

Choking	Disease duration ≤ 1 year (n=22)	Disease duration >1 year (n=18)	*P- value*
Absent [N (%)]	22 (100%)	12(66.7%)	0.003* (FET)
Present [N (%)]	0(0%)	6(33.3%)	

Fisher Exact test was used

Patients with a number of relapses >2 relapses have a significantly higher incidence of choking than patients with a number of relapses ≤ 2 relapses ($P = 0.028$). (Table 5) illustrates these data.

Table 5: Effect of a number of relapses throughout disease duration on choking

Choking	Number of relapses ≤ 2 relapses (n= 23)	Number of relapses >2 relapses (n=17)	*P- value*
Absent [N (%)]	22(95.7%)	12(70.6%)	0.028* (X2)
Present [N (%)]	1(4.3%)	5(29.4%)	

X^2= Chi-Squared test was used

Patients with EDSS >3 have a significantly higher incidence of choking than patients with EDSS ≤ 3 (P = 0.001). (Table 6) shows the previous data.

Table 6: Effect of EDSS on choking

Choking	EDSS ≤ 3 (n= 24)	EDSS >3 (n=16)	*P- value*
Absent [N (%)]	24(100%)	10(62.5%)	0.001** (FET)
Present [N (%)]	0(0)	6(37.5%)	

Fisher Exact test was used

There was no statistically significant correlation between FEES findings and either patients' age, age of the patient at disease onset, disease duration, number of relapses throughout disease duration, and EDSS score ($p > 0.05$). These data are summarized in (Table 7).

Table 7: Correlation between FEES regarding patients' characteristics

Patient characteristics	FEES	
	(r) coefficient (spearman)	*P-value*
Age	0.073	0.653
Age of disease onset in years	0.248	0.123
Disease duration in years	-0.206	0.201
Number of relapses throughout disease duration	-0.164	0.312
EDSS	-0.146	0.313

Spearman non-parametric correlation

DISCUSSION

In the present study, FEES revealed that 50 % of RRMS patients had no residue, 35% had mild residue and 15% had moderate residue on The Yale Pharyngeal Residue Severity Rating Scale. We also found that 15% of the patients had choked with penetration, but there was no aspiration or nasal regurgitation. It is important to clarify that the presence of choking with penetration means that these patients still have protective cough reflex and preserved laryngeal sensations.

In accordance with our findings, Poorjavad and his colleagues conducted a study to evaluate the spread of swallowing disorders in MS patients. The authors used Northwestern Dysphagia Patient Check Sheet for the assessment of swallowing functions in 101 MS cases. A total of 32 cases (31.7%) were found to have dysphagia. Notably, aspiration, oral stage disorders, and pharyngeal delay were observed in 6.9%, 5%, and 1% of patients, respectively[11].

Likewise, Guan et al. conducted a meta-analysis on 15 articles. About 4.510 cases were enrolled in the analysis, and dysphagia was detected in 36% of the tested population[17].

Similarly, Alali et al. included a total of 103 adults with MS to evaluate the same previous perspective. About 38% of adults with MS reported having swallowing problems that resulted in various physical and social consequences[12]. Furthermore, the previous findings were similar to reports by other previous two studies conducted in the late nineties[18,19].

Other reports showed a higher frequency of dysphagia among patients with MS. For example, Fernandes et al. assessed the frequency of dysphagia in patients with MS. In their study, 120 MS patients underwent a functional assessment of swallowing. Dysphagia was reported in 90% of patients[20].

Likewise, Alfonsi et al. made an electrophysiological study of swallowing in MS and found that 92% of patients showed at least one electrophysiological abnormality[21]. The exact causes of such difference between our findings and the above-mentioned studies are not clear. However, this difference could be explained by the variations in the characteristics of the included patients, the difference in sample size may be another cause. Also, the method of dysphagia assessment is an important cause as the electrophysiological study of swallowing is totally different from FEES. While FEES gives informative data about the presence and degree of residue, aspiration, penetration, choking, and nasal regurgitation, the electrophysiological study of swallowing (EPSS) measures different items like the duration of suprahyoid/submental muscle EMG activity and the duration of the laryngeal–pharyngeal mechanogram.

In terms of the association between swallowing impairment and clinical characteristics of the patients, we found that there was a statistically significant difference between patients without residue, with mild residue, and with moderate residue regarding the patient age at onset of the disease. Older age at the onset of the disease was associated with more swallowing impairment. On the contrary, Poorjavad et al. revealed no significant association between dysphagia and the age of the patient at MS onset[11].

The present study revealed no statistically significant difference between the three groups of FEES findings in terms of disease duration. On the other hand, patients with a disease duration >1 year have a significantly higher incidence of choking than patients with disease duration ≤ 1 year.

Similar to our findings, Poorjavad et al. reported that patients with dysphagia had longer MS duration ($p = 0.031$) and more cerebellar impairment ($p = 0.04$) compared with non-dysphagic patients[11].

The present study revealed no statistically significant difference between the three groups of FEES findings in terms of number of relapses. On the other hand, patients with number of relapses > 2 relapses throughout disease duration have significantly higher incidence of choking than patients with number of relapses ≤ 2 relapses. This can be explained by that MS is usually getting severe or debilitating over time. The swallowing impairment may get worse with each disease attack.

Likewise, Fernandes et al. reported that patients with more than two relapses had a significantly higher incidence of dysphagia[20].

The present study showed no statistically significant difference between the three groups of FEES findings in terms of EDSS. On the other hand, patients with EDSS > 3 have a significantly higher incidence of choking than patients with EDSS ≤3.

Similar to our findings, Clavé Civit and his colleagues studied 23 patients suffering from MS. Patients with dysphagia were found to have significantly higher EDSS scores[22]. Moreover, Poorjavad et al. reported that the prevalence of dysphagia was significantly higher in patients with more severe neurological disabilities measured by EDSS ($p = 0.04$)[11].

In addition, Abraham et al. studied 525 MS cases, and about 40% of the included sample reported dysphagia. Patients reporting dysphagia tended to have greater impairment in mental, cerebellar, and brainstem functions compared to patients without swallowing problems[19].

Fernandes et al. reported that, regarding the EDSS, higher scores were associated with severe dysphagia[20]. De Pauwa et al. (2002) recruited 300 consecutive MS patients in their study, and they reported that permanent and transient swallowing problems were reported in 24 and 5% of cases respectively. Permanent dysphagia tended to arise in mildly impaired patients (EDSS 2 – 3), while its prevalence increased together with a rising disability to reach 65% in the most severely disabled subjects (EDSS 8 – 9).

Our study has some limitations; first, it is a single-center study. Secondly, the included sample size was relatively small, and the study didn't include other MS types such as primary or secondary progressive MS; also, the actual dynamics of the swallowing were not assessed by FEES. Hence, more studies including more cases from different neurological centers should be conducted soon; also, an assessment of swallowing of MS patients using videofluoroscopy should be planned, finally, single research containing EPSS, and FEES may give another idea about swallowing impairment in MS patients.

CONCLUSION

Our findings highlighted that about half of the RRMS patients are suffering from swallowing difficulties, with variable degrees. Disease duration >1 year, a number of relapses >2 relapses, and EDSS >3 are significant risk factors for choking in MS patients. This might encourage neurologists to properly assess the swallowing function in MS cases especially if they have a high EDSS score and long disease duration. Nevertheless, further large-scale studies are still needed to confirm our findings. Additional studies are recommended for better comprehension of swallowing impairment in MS and giving information on the expansion of evaluation methods and management.

CONFLICT OF INTERESTS

There are no conflicts of interest.

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