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ORIGINAL ARTICLE

Prevalence of dysphagia in a consecutive cohort of subjects with MS using fibre-optic endoscopy

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Abstract

Introduction Multiple sclerosis (MS) refers to chronic inflammation of the central nervous system including the brain and spinal cord. Dysphagia is a symptom that represents challenges in clinical practice. The aim of the present study was to evaluate the prevalence of dysphagia in an Italian cohort of subjects with MS using the Dysphagia Outcome Severity Score (DOSS), based on fibre-optic endoscopy, and determine factors that correlate with the presence of swallowing problems.

Matherials and Methods Data were collected in a multicentre study from a consecutive sample of MS patients, irrespective of self-reported dysphagia. The study included 215 subjects. Possible scores for DOSS range from 7 to 1, with 7 indicating normal swallowing.

Results One hundred twenty-four (57.7%) subjects demonstrated abnormal swallowing and 57 (26.5%) of these had swallowing problems that required nutrition/diet modifications when evaluated objectively with fibre-optic endoscopy. Subjects with dysphagia were more severely disabled and more often had a progressive form of MS, compared to MS subjects with normal swallowing. In subjects with EDSS, < 4, 8 (13.3%), had a DOSS < 4. Seventy-five percent of subjects older than 60 years of age had dysphagia.

Conclusion In this sample of MS patients, more nearly 60% showed swallowing problems.

Keywords Multiple sclerosis · Dysphagia · FEES · DOSS · Deglutition · Deglutition disorders

Introduction

Swallowing problems can be part of the array of symptoms related to multiple sclerosis (MS). A recent systematic review and meta-analysis on the prevalence of dysphagia in MS showed different rates according to the measures utilised, varying from 31% for subjective measures to 81% with the use of instrumental evaluations [1].

Dysphagia has been reported in patients with mild MS (17%), and appears to increase in frequency as the level of disability worsens, with a reported prevalence of 65% in severely disabled subjects [2–4], scored using the Expanded Disability Status Scale (EDSS) [5].

The gold standard in swallowing assessment includes the videofluorographic swallowing study and fibre-optic endoscopic evaluation of swallowing (FEES) [6, 7]. FEES is a technique for the evaluation of pharyngeal swallowing in different neurological diseases, such as Parkinson disease, acute stroke, MS, amyotrophic lateral sclerosis and Alzheimer's disease [8]. The majority of these studies were performed in relatively small populations [9, 10]. The Dysphagia Outcome Severity Scale (DOSS) is commonly utilised for grading results of the FEES [11].

To our knowledge, only a few studies have evaluated the presence of dysphagia in MS using FEES. In particular, Calcagno et al. evaluated swallowing in 143 consecutive patients with primary and secondary progressive MS.

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Evaluations included both the FEES scored using a 3-point scale and a speech pathology assessment that included a preliminary interview to establish if the patient had experienced any subjective symptoms related to dysphagia, an evaluation of laryngeal elevation and direct observation of morphology, sensibility and motility of the lips and tongue. A water swallow test had been carried out. Dysphagia was diagnosed in 49 subjects (34.3%) with a significant correlation with illness severity [12]. In a second study, Alfonsi and colleagues evaluated dysphagia in 26 subjects with MS and FEES was graded using the penetration-aspiration scale (PAS), ranging from 1 (normal) to 8 [13]. FEES was altered in 14/26 patients (53.8%), with a score of 2 in 3 subjects (11.5%) and 0 in 12 subjects (46.2%). The most frequent alterations were related to prolongation of both the propulsive oral and the pharyngeal phases. These results confirmed that laryngopharyngeal motility dysfunction with a prolonged interval between the oral and pharyngeal phases of swallowing is the primary cause of dysphagia in MS [13].

The aim of the present study was to evaluate the prevalence of dysphagia in an Italian cohort of subjects with MS using the DOSS based on fibre-optic endoscopy and to assess whether any clinical characteristics related to MS correlate with DOSS scores.

Material and methods

Subjects

Study participants were a consecutive sample of patients followed at four Italian MS clinics (Santa Lucia Hospital Rome, ASL3 Genovese Genova, IRCCS Mondino Foundation Pavia, University of Catania). Inclusion criteria were a diagnosis of MS according to the McDonald revised criteria [14], a minimum age of 18 years, no relapses in the last 3 months and no treatment or training for dysphagia in the last 3 months prior to study entry. The only exclusion criterion was the presence of diseases other than MS influencing swallowing function.

Instruments

FEES is an instrumental evaluation in which a flexible endoscope is introduced transnasally in the patient's pharynx providing a view of the laryngeal and pharyngeal structures and their functions [15]. The Dysphagia Outcome Severity Score (DOSS) is a scale used for assigning a score to the fibre-optic endoscopy findings, ranging from 7 to 1. Level 7 indicates normal swallowing. Level 6 is consistent with full oral nutrition with a normal diet, although implies some functional limitations (such as extra time for meals, mild oral and pharyngeal delay, retention or trace epiglottal undercoating), for which the subject independently and spontaneously compensates. Levels 5 to 3 indicate the need for modified diet and/or independence and levels 2 and 1 exclude oral nutrition.

The Expanded Disability Status Scale (EDSS) is used to quantify disability in MS and to monitor changes in the level of disability overtime and is widely used in clinical trials in MS [5]. EDSS scores range from 0 to 10 in 0.5 unit increments with a higher score representing a higher level of disability. Scoring is based on a neurological examination by a qualified medical specialist.

Procedure

Subjects consecutively presenting at MS clinics provided written informed consent to participate in the study. A standardised FEES protocol was used to examine the mouth, teeth, pharyngeal velum, tongue, pharynx, larynx and voice quality, both for morphological and functional abilities [16]. Examiners were blinded to subjects' medical history, disease severity and the presence of self-reported swallowing symptoms. The study was approved by the Ethics Committees of centers involved in the study (PI CE: AG4-PROG259-157).

Statistical analyses

Patients were divided into two groups, based on the presence of dysphagia: DOSS = 7 normal swallowing, DOSS < 7 dysphagia). Parametric tests were used to compare age and disease duration (unpaired t tests to compare groups) between patients with and without dysphagia according to DOSS levels. Non-parametric tests were used to assess differences between groups on EDSS, (Wilcoxon-Mann-Whitney rank sum test for two groups). Categorical data were compared using the chi-square test. A two-tailed p value of < 0.05 was considered statistically significant. Means, standard deviations (SD) or medians and ranges for continuous variables (age, EDSS, number of years since diagnosis and first symptoms) and percentages for all other categorical factors were analysed. To assess differences between patients with and without dysphagia, each demographic (age, gender) and clinical characteristic (EDSS, time since diagnosis and since first symptom and disease stage) was individually tested with a chi-square test (gender), Student's t test for independent data (age) or the non-parametric Mann-Whitney test (EDSS, time from diagnosis, time from first symptom). Characteristics found to be significantly associated with the presence of dysphagia were analysed in a multivariate logistic regression with dysphagia (yes/no) as the dependent variable. SPSS software version 17 for Windows (SPSS, Chicago IL, 2002) was used for the statistical analyses.

Results

Two hundred and nineteen consecutively presenting patients with MS were initially enrolled in the study. Four subjects were subsequently excluded from the analysis due to incomplete examinations. The final sample included 215 subjects: 82 males (38%) and 133 females (62%), ratio F:M = 1.6:1, with a mean age of 50 years (SD ± 11.9) and a mean disease duration of 15.1 (SD ± 10.2 years). The mean EDSS was 5.4 (SD ± 2.4, range 0–9.0). One hundred three subjects (47.9%) had a relapsing-remitting form of MS, 103 (47.9%) a secondary-progressive form and 9 (5.6%) a primary progressive disease course (Table 1). No significant centre effect was observed for the distribution of clinical variables.

Figure 1 shows the results of FEES findings scored according to DOSS levels. Ninety-one subjects (42.3%) had a DOSS score of 7 (normal swallowing). Patients with dysphagia had more severe disability, evaluated by EDSS ($p < 0.001$), more frequently had a progressive form of the disease (primary progressive and secondary progressive) ($p < 0.002$) and a longer disease duration ($p < 0.002$), compared to patients with normal swallowing. No statistical differences were found for age and gender between the groups (Table 1). The multivariate analysis demonstrated a correlation between dysphagia and disease course and level of disability but not with gender, age or disease duration, although age trended towards statistical significance (0.07).

In a post hoc analysis, out of 44 subjects over the age of 60, 33 (75%) had swallowing problems.

In patients with EDSS < 4, the typical MS outpatient, out of 60 subjects, 25 (41.7%) had a DOSS < 7 with no differences for demographic characteristics compared to subjects with a DOSS < 7. In this group, 17 subjects had a DOSS = 6 and 8 subjects had DOSS = 5, representing significant swallowing problems. This subgroup had no differences in demographic characteristics compared to subjects with DOSS = 6 or 7. Considering 114 patients with a progressive disease course (PP and SP), two-thirds of subjects had swallowing problems, of whom 27 subjects showed DOSS = 6.

Discussion

The prevalence of dysphagia in the present large consecutive series of MS outpatients, based on an objective assessment, irrespective of the presence of reported symptoms, is consistent with data previously published, indicating that dysphagia is a relatively frequent symptom [3, 17–19].

To our knowledge, this represents the first attempt to analyse an unselected consecutive sample of MS patients for swallowing disorders, reporting the distribution of DOSS in a large MS population.

FEES allows direct three-dimensional visualisation of the pharynx and larynx before and after swallowing and is a procedure that can be repeated when necessary [20]. A recent systematic review [1] reported variability in the prevalence of dysphagia in patients with MS that varies according to the diagnostic methods utilised, specifically subjective vs. objective screening. Only 4 studies used objective measures and the sample size varied from 18 to 120 MS patients. Despite the high heterogeneity of results from various studies (due to diagnostic methods, disease duration and disability severity), comparing subjective and objective methods, the authors concluded that more than one-third of subjects with MS experience swallowing difficulties [1].

Table 1 Demographic and clinical characteristics of subjects with normal swallowing and subjects with dysphagia

	Total subjects ($n = 215$)	Subjects with no dysphagia ($n = 91$; 42.3%)	Subjects with dysphagia ($n = 124$; 57.7%)	p value
Age in years (SD)	50 (11.86)	47.49 (11.89)	51.98 (11.64)	0.1
Gender F (%)	133 (61.9)	60 (65.9)	73 (58,8)	0.32
Mean EDSS (SD, range)	5.39 (2.37, 0–9.5)	4.71 (2.40, 1–9)	5.91 (2.22, 1–9.5)	0.001*
MS type				
RR (%)	103 (47.9)	55 (60.4)	48 (38.7)	< 0.001*
SP (%)	103 (46.5)	34 (37.4)	69 (55.6)	
PP (%)	9 (5.6)	2 (2.2)	7 (5.6)	
Mean disease duration in years (SD)	15.07 (10.17)	14.03 (10.66)	15.85 (9.75)	0.001

Subjects were divided into two groups based on the presence of dysphagia at the time of clinical evaluation (DOSS < 7)

EDSS, Expanded Disability Status Scale; *RR*, relapsing remitting; *SP*, secondary progressive; *PP*, primary progressive; *indicates statistical significance on multivariate analysis

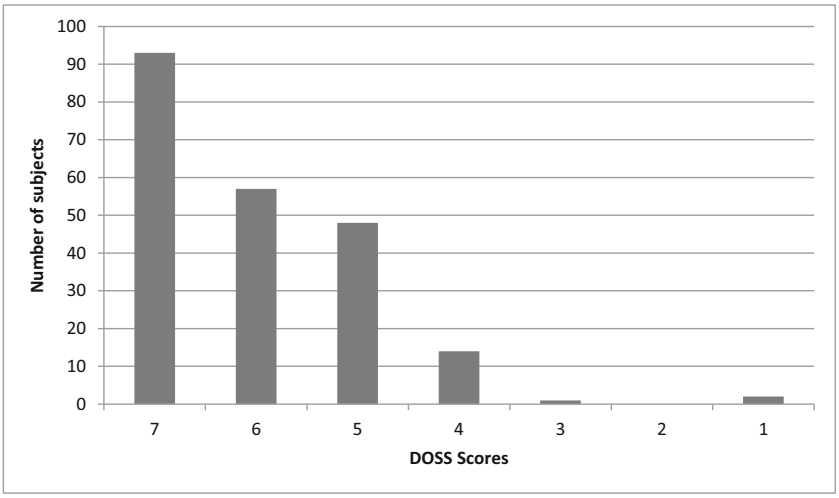

Fig. 1 Distribution of DOSS scores. DOSS, Dysphagia Outcome Severity Score.

DOSS- Dysphagia Outcome Severity Score

Consistent with previous findings, there was a significant correlation between DOSS and EDSS [3]. In the current sample, 25 out of 114 subjects (21.9%) with dysphagia had an EDSS score from 1 to 3.5, confirming the relatively high frequency of dysphagia even in mild forms of MS. Further, considering subjects with EDSS < 4, almost 42% had swallowing problems, underlining the importance of evaluating dysphagia even in subjects with lower levels of disability. Importantly, 8 subjects out of 60 with lower EDSS (13.3%) actually had moderate swallowing problems. Moreover, among different clinical forms of MS, progressive forms (PP and SP) were more frequently associated with severe dysphagia, while the relapsing-remitting form presented more often mild to moderate dysphagia [12]. These results are consistent with the report from Calcagno where a similar frequency of dysphagia was reported, although the study included exclusively progressive forms of MS where the level of disability was higher than in the current study [12]. The present study confirmed a significant correlation between swallowing dysfunction and progressive MS with more advanced disability.

Conclusion

The present study confirmed dysphagia in nearly 60% of subjects. Dysphagia is significantly correlated with progressive MS and a higher level of disability. Swallowing problems are relatively frequent in subjects with lower levels of disability (41.7%) and are quite frequent in older subjects (75%).

Given that very serious swallowing difficulties were detected in 8 subjects with an EDSS < 4, FEES should not be used across patients, due to the fact that it may be considered too invasive as a screening tool. Although the presence of undiagnosed swallowing difficulties, symptoms that can put the patient at risk for serious complications, does underline the necessity for routine screening in MS outpatients with the use of non-invasive means for detecting swallowing problems. The FEES is recommended as a standard screening method in subjects with a higher level of disability who appear to be more at risk for the presence of swallowing problems and in individuals over 60 years of age.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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