# INPLASY

## Diagnostic accuracy of self-reported dysphagia questionnaires in multiple sclerosis: Meta-analytic study

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## ADMINISTRATIVE INFORMATION

## INTRODUCTION

Review question / Objective The population (P) are patients diagnosed with multiple sclerosis (MS) who are included in the self-assessment (I) of dysphagia, which is compared to (C) the objective assessment procedure or other validated dysphagia assessment. The objective (O) of this meta-analytic study is to define and assess the specificity and sensitivity of these questionnaires through diagnostic accuracy procedures.

Rationale Diagnosis of dysphagia varies according to subjective and objective assessment tools from 37.21% to 58.47%, and the total prevalence of dysphagia is 43.33%. According to the systematic review of nine self-assessment dysphagia tools in progressive neurodegenerative diseases, there is a need for evaluation, validation, and greater availability and routine use of diagnostic tools for dysphagia. Thus, meta-analytic diagnostic accuracy procedures for dysphagia in MS will be made to identify the best tools for diagnosing dysphagia in MS and easy application of these assessment tools due to the need for regular follow-up assessments according to the nature of MS. Also, an additional reason why regular follow-up assessments are necessary is the great variety of dysphagia symptoms during the course of the disease.

Condition being studied Multiple sclerosis (MS) is an inflammatory autoimmune degenerative central nervous system (CNS) disease. It causes demyelination and axonal loss with the formation of plaques. The clinical course of the disease is heterogeneous and associated with demyelization and plaque localization. One of the common disabilities is dysphagia.
Dysphagia in earlier stages of MS (EDSS<6) is often an undiagnosed clinical symptom, mostly because it is present in the form of subclinical dysphagia in 30% of patients with mild or moderate MS. Pharyngeal phase of swallowing is mostly affected in patients with mild to moderate MS.

Dysphagia is the number one cause of fatal outcomes in MS. Dysphagia prevalence in severely disabled patients ranges from 65% (EDSS 8-9) to 95% (EDSS 9.5). It is generally accepted that dysphagia occurs in the advanced phase of MS. However, the literature showed that the prevalence of dysphagia is 33-43% in early phases of MS, while it is diagnosed in 30% of patients with relapse-remitting multiple sclerosis (RRMS), mostly in later phases. The onset of dysphagia is typical for advanced phases of MS, but according to some researchers it can occur in early phases with EDSS score 2-3 with prevalence of 9,95 to 20%. There is a presence of heterogeneity in the previous prevalence study, and a recent meta-analysis showed heterogeneity in the geographical location of patients, according to different diagnostic procedures and undefined disease severity score (EDSS).

## METHODS

**Search strategy** In the literature search, a combination of the following keywords will be used: Dysphagia, swallowing, oropharyngeal dysphagia, neurogenic dysphagia, multiple sclerosis, demyelination disease, neurodegenerative disease, self-assessment questionnaires.

**Participant or population** People with diagnosed MS, aged 18 and above. The exclusion criteria are the presence of other neurological conditions that are not connected to MS.

**Intervention** Application of any self-assessment questionnaire for dysphagia in patients with MS.

**Comparator** Comparison to other dysphagia assessment procedures that are considered standard in clinical assessment.

**Study designs to be included** Original scientific papers, cross-sectional studies, diagnostic accuracy studies, and systematic reviews will be included.

**Eligibility criteria** The papers included for full review need to have information on the development or validation process of subjective or self-reported dysphagia questionaries, population description and the results. All assessment tools included in the review need to be used in the MS population. The main inclusion criteria for the population are the presence and diagnose of MS.
All articles published in English language will be assessed for full-text analysis, without any time-based restrictions.

Exclusion criteria will be case studies and case reports, non-validated assessment procedures, unclear diagnostic methods.

**Information sources** Published papers will be searched in scientific databases: Ovid, Scopus, and Web of Science.

**Main outcome(s)** The main outcome is to identify which self-assessment, subjective questionnaire has good sensitivity and specificity in comparison to the index test and define its diagnostic accuracy.

**Additional outcome(s)** Additional outcomes will be to define sensitive and accurate dysphagia self-assessment questionnaire that can be used as the standard in clinical screening procedures and in prevalence studies.

**Data management** Two reviewers will individually screen the papers according to the title and abstract of all articles included in the review. In case there is no consensus of two reviewers, the third reviewer will decide on the inclusion of the screened papers. After consensus for paper eligibility, full-text articles will be observed.
Data will be extracted using a developed extracting form tailored to the needs of this meta-analytic study. Two reviewers will independently extract all data to avoid biases. One reviewer will extract all the data, and the other one will assess it independently to avoid biases. After data extraction, all forms will be analyzed and summarized by the whole team of authors to avoid disagreements. Abstracted data will include demographic information, information about the diagnostic procedures, statistical results extracted from validation, and positive and negative aspects of assessment elaborated in the article. If any uncertainties occur, the reviewers will contact the study authors directly.

**Quality assessment / Risk of bias analysis** Each study included in the meta-analytic process will be assessed for possible bias using the QUADAS-2 protocol. For each study included, all tool domains will be analyzed from the extracted data. The risk of bias will be assessed by two reviewers independently to avoid further biases. In case of a disagreement between reviewers, there will be a discussion about the assessment, or a third author will be asked to assess the study independently. The assessment of study quality will be presented graphically.

**Strategy of data synthesis** Collected data will be analyzed using qualitative and quantitative

approaches. Qualitative analysis will be made for every diagnostic tool included in the meta-analytic study in a narrative form including tables and figures to aid in data presentation where appropriate. Meta-analytic procedures will be made in quantitative analysis using the STATA19 Statistical Software

Reliability and validity, including sensitivity, specificity, positive predictive value and negative predictive value and likelihood ratios, will be extracted directly from the source papers. If this is not possible, values will be calculated from the data provided. Positive and negative likelihood ratios, diagnostic odds ratios and 95% confidence intervals will be calculated. ROC curve will be used to display the data with the overall AUC (area under the curve) to indicate the overall performance of the test.

**Subgroup analysis** None.

**Sensitivity analysis** None.

**Language restriction** Articles published in English language will be assessed for full-text analysis.

**Country(ies) involved** Croatia.

**Keywords** Multiple sclerosis, dysphagia, self-assessment, questionnaire, diagnostic accuracy, sensitivity.

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