Dysphagia Screening in Parkinson's Disease. A diagnostic accuracy cross-sectional study investigating the applicability of the Gugging Swallowing Screen (GUSS)

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Abstract

Background: Simple water-swallowing screening tools are not predictive of aspiration and dysphagia in patients with Parkinson's Disease (PD). We investigated the diagnostic accuracy of a multi-texture screening tool, the Gugging Swallowing Screen (GUSS) to identify aspiration and dysphagia/penetration in PD patients compared to flexible endoscopic evaluation of swallowing (FEES).

Methods: Swallowing function was evaluated in 51 PD participants in clinical ‘on-medication’ state with the GUSS and a FEES examination according to standardized protocols. Inter-rater reliability and convergent validity were determined and GUSS- and FEES-based diet recommendations were compared.

Key Results: Inter-rater reliability of GUSS ratings was high ($r_s = 0.8; p < 0.001$). Aspiration was identified by the GUSS with a sensitivity of 50%, and specificity of 51.35% (PPV 28%, NPV 73%, LR+ 1.03, LR- 0.97), dysphagia/penetration was identified with 72.97% sensitivity and 35.71% specificity (PPV 75%, NPV 33.33%, LR+ 1.14, LR- 0.76). Agreement between GUSS- and FEES-based diet recommendations was low ($r_s = 0.12, p = 0.42$) with consistent NPO (Nil per Os) allocation by GUSS and FEES in only one participant.

Conclusions and Inferences: The multi-texture screening tool GUSS in its current form, although applicable with good inter-rater reliability, does not detect aspiration in PD patients with acceptable accuracy. Modifications of the GUSS parameters “coughing,” “voice change” and “delayed swallowing” might enhance validity. The GUSS’ diet recommendations overestimate the need for oral intake restriction in PD patients and should be verified by instrumental swallowing examination.

KEYWORDS
aspiration, dysphagia, FEES, Gugging Swallowing Screen, Parkinson's disease
1 | INTRODUCTION

Neurogenic dysphagia is highly prevalent in patients with Parkinson’s disease (PD) with significant consequences for nutritional status, quality of life, and outcome.1 Dysphagic symptoms may occur in the early stages of the disease,2 however, self-perception of dysphagia might be limited in PD.3 Typical dysphagia-related findings include oral stage impairments such as dyscoordinated tongue movements, impaired masticatory function, prolonged oral transit time (OTT), and pharyngeal stage impairments including residues, penetration, aspiration, and pharyngoesophageal dysmotility.4,5 The pathology behind aspiration seems to be a combination of pharyngeal sensory and motor impairments. Additionally, many PD patients have problems with generating sufficient expiratory flow and subglottic pressure to cough effectively.6

A gold standard method to identify aspiration in PD patients is flexible endoscopic evaluation of swallowing (FEES).7,8 as it detects dysphagia in more than 50% of subjectively asymptomatic patients.3 However, instrumental swallowing assessment is associated with costs and burden for the patient and availability depends on clinical settings. This emphasizes the need for a dysphagia screening tool with acceptable sensitivity and specificity to identify aspiration to determine the necessity for continued further instrumental assessment throughout the progression of the disease.

To date, no PD-specific dysphagia screening tool exists. Simple water-swallowing tests, measurements of volume and speed while drinking fluids and swallowing questionnaires are not predictive of aspiration in PD patients.9,10 Considering the complex pathophysiology behind dysphagia in PD and the increase of impairments with swallowing solid food,2 dysphagia and aspiration could be noticeable earlier in this patient group when multi-texture screening tools are applied. The Gugging Swallowing Screen (GUSS),11 a widely used multi-texture tool developed and validated for acute stroke patients, might be applicable. The GUSS includes successive evaluation of saliva swallows and three different bolus textures (semi-solid, liquid, solid) based on the occurrence of established signs of dysphagia and aspiration. A sum score is calculated and interpreted in terms of aspiration risk and dysphagia risk, and diet recommendations are provided. Sensitivity (≥96%) and specificity (≥50%) of the GUSS to detect aspiration risk in acute stroke patients and good inter-rater reliability (κ = 0.84) have been established.11,12 Diet recommendations derived from the GUSS sum score were found to be more conservative than those determined by a FEES-based assessment rated with the Fiberoptic Endoscopic Dysphagia Severity Scale (FEDSS).13 with the GUSS overestimating the need for a non-oral diet (Nil Per Os - NPO) and tube feeding in stroke patients.12

The objective of this study was to investigate the applicability of the GUSS in patients with PD by:

- establishing inter-rater reliability when the GUSS is applied in this patient group
- determining the diagnostic accuracy of the GUSS to identify aspiration and dysphagia/penetration compared to a gold standard (FEES)
- investigating the agreement between GUSS-based and FEES-based diet recommendations

2 | MATERIAL AND METHODS

2.1 | Ethical approval

Data collection in this prospective cross-sectional study was conducted at the University Medical Center Hamburg-Eppendorf between January 16, and January 31, 2019. It was approved by the local ethics committee of the Medical Council Hamburg. Written informed consent was obtained from all participants.

2.2 | Participants

A cohort of 56 consecutive patients with confirmed PD diagnosis14 were recruited who either attended a regular consulting at the movement disorders outpatient clinic or were inpatients at the Medical Center’s neurological ward. Patients with atypical or secondary Parkinson syndromes and diseases associated with dysphagia were excluded, as well as patients with cognitive limitations that would preclude adequate comprehension of instructions. 51 participants were included in the study, and 5 participants were excluded on the basis of the exclusion criteria.

The participants’ motor functional status was assessed with the new revised Movement Disorder Society version of the Unified Parkinson’s Disease Rating Scale (MDS-UPDRS part III).15 Hoehn and Yahr stage (H&Y)16 was ascertained; for data analysis, H&Y 2.5 was included in stage 2 and H&Y 3.5 was included in stage 3. The cognitive status was assessed with the Montreal Cognitive Assessment (MoCA).17 Participants were interviewed with an inform<br>
TABLE 1 Characteristics of participants (N = 51 PD patients)

| Characteristic               | Mean ±SD or N (%) |
|------------------------------|-------------------|
| Age (years)                  | 69.8 ± 9.8        |
| Gender (f/m)                 | 18/33             |
| Disease duration             | 10.3 ± 9.2        |
| Hoehn & Yahr                 |                   |
| Stage 1                      | 1 (1.96%)         |
| Stage 2                      | 27 (52.94%)       |
| Stage 3                      | 15 (29.41%)       |
| Stage 4                      | 7 (13.73%)        |
| Stage 5                      | 1 (1.96%)         |
| DBS                          | 10 (19.61%)       |
| MDS-UPDRS motor score        | 27.9 ± 14.2       |
| MOCA score                   | 25.2 ± 4.2        |
| FOIS                         | 6.6 ± 0.8         |
| History of pneumonia         | 0                 |
| Current diet modifications   | 12 (23.53%)       |

Abbreviations: DBS, Deep Brain Stimulation; MDS-UPDRS, Movement Disorder Society-sponsored revision of the unified Parkinson’s disease rating scale; MOCA, Montreal cognitive assessment; FOIS, Functional Oral Intake Scale.

The GUSS was conducted by two Speech Language Pathologists (SLPs) according to the standardized stepwise GUSS protocol including assessment of saliva and three different bolus textures (see below) in increasing volumes. 

FEES was carried out by experienced (>15 years) otolaryngologists using a 2.6-mm-diameter high-definition rhino-laryngoscope (ENT-V3, Olympus Medical Systems Corp., Tokyo, Japan) according to the protocol of the FEDSS.

During both examinations, participants were given the same three standardized test boluses in a fixed order:

1. **Semi-solid**: 3× 1/2 teaspoons of thickened water (IDDSI level 3)
2. **Liquid**: 5, 20, and 50 mL of water (IDDSI level 0) using a straw. Patients were asked to drink the water quickly but not as fast as possible
3. **Solid**: 3 pieces of bread with butter (ca. 30 × 30 mm, weight 7 g, IDDSI level 7 minus, easy to chew)

All consecutive test boluses and volumes were administered to allow valid blinded retrospective re-rating unless the patient refused to continue or the examiner decided to terminate the examination. All examinations were recorded on video (MediCap USB300 MediciCapture, Plymouth Meeting; PA, USA) and evaluated by a second rater (rater #2) afterward in order to establish inter-rater reliability of data collection in identical assessment situations.

### 2.4 Data analysis

The GUSS examinations were rated by 2 independent and blinded SLPs according to the annotated version of the GUSS and the GUSS manual (https://www.dysphagie-trapl.at/guss-formular-deutsch/). Scores in the GUSS subtests were assigned based on the presence of signs of dysphagia as defined in the GUSS protocol: (i) reduced vigilance, (ii) weak voluntary and reflexive coughing, (iii) drooling, (iv) absent or delayed swallowing, and (v) voice change. The occurrence of one of these indicators led to the termination of the scoring and assignment of the final GUSS sum score. This sum score was interpreted in terms of aspiration risk, dysphagia risk, and diet recommendations as described in the manual.

FEES examinations were rated by two independent and blinded otolaryngologists (>15 years of experience). The occurrence of penetration and aspiration was rated with the Penetration-Aspiration Scale (PAS). In setting the cutoff scores for aspiration and dysphagia/penetration, we replicated procedures of the previous GUSS validation studies. "Aspiration" was defined as a GUSS cutoff score of ≤14 and a PAS score of ≤6. "Dysphagia/penetration" was defined as a GUSS cutoff score of ≤19 and a PAS score of ≥3. Instead of the terms “aspiration risk” and “dysphagia risk” used in the GUSS, we used “aspiration” and “dysphagia/penetration” in the following analyses to standardize reporting of the results.
Diet recommendations were derived from the FEES examination using the FEDSS\(^{13}\) score. The FEDSS scaling was slightly adjusted as shown in Table 2 to allow comparison with the GUSS’ recommendations.

### 2.5 Statistical procedures

Quantitative data describing participants’ characteristics were calculated with mean, standard deviations (SD), and percentages. Effects of H&Y stage and gender on GUSS scores were analyzed with univariate analysis of variance (ANOVA), effects of age, disease duration, motor functional status (MDS-UPDRS part III), and cognitive status (MoCA) were analyzed by multiple linear regression analysis.

Inter-rater reliability of the two independent GUSS ratings was established with the Spearman rank correlation coefficient (r\(_S\)). Inter-rater agreement of the FEES ratings was determined by Bravais-Pearson’s correlation coefficient.

Accuracy of the GUSS (compared to FEES) to detect aspiration and dysphagia/penetration was analyzed with a 2 × 2 contingency table and the following diagnostic characteristics were extracted: sensitivity [TP/(TP+FN)], specificity [TN/(TN+FP)], positive predictive value [TP/(TP+FP)], negative predictive value [TN/(TN+FN)] (TP=true positive, FP=false positive, TN=true negative, FN=false negative), and likelihood ratios [LR+ = sensitivity/1-specificity; LR− = 1-sensitivity/specificity]. Acceptable sensitivity was defined as ≥80%, and acceptable specificity was defined as ≥70%.\(^{10}\) A moderate change in pre- to post-test probability defined as LR+ ≥2 and LR- ≤0.2 was considered acceptable.\(^{20}\) Exact Clopper Pearson confidence intervals (95% CI) were used for sensitivity/specificity, log method for likelihood ratios,\(^{21}\) and standard logit CIs\(^{22}\) for predictive values.

Agreement between GUSS and FEDSS diet recommendations was analyzed with the Spearman rank correlation coefficient, and other comparisons with respect to diet recommendations were done descriptively.

All statistical tests were two-tailed, and a priori significance levels were set at \(p < 0.05\). Data were analyzed with the statistical software package SPSS, version 24.0 (IBM USA).

## 3 RESULTS

### 3.1 GUSS results

The group had a mean GUSS sum score of 14.2 (SD = 5.9; 4–20). The data revealed no effect of H&Y stage [\(F(4,46) = 1.532, p = 0.209\)] and gender [\(F(1,49) = 1.823, p = 0.183\)] on the GUSS scores and no systematic relationship between GUSS scores and age [\(F(1,49) = 1.880, p = 0.353, R^2 = 0.018\)] and cognitive status (MoCA scores) [\(F(1,49) = 0.567, p = 0.455, R^2 = 0.011\)]. However, a relationship between GUSS scores and MDS-UPDRS motor scores \([\beta = −0.135, t = 2.409, p = 0.20\]) and GUSS scores and disease duration \([\beta = −0.224, t = 2.218, p = 0.32\]) was found, so that participants with more pronounced motor symptoms (higher MDS-UPDRS III scores) and longer disease duration had lower GUSS scores.

Only 15 participants (29%) passed all conditions of the GUSS, and dysphagia symptoms (GUSS sum scores ≤19) were found in 36 participants (71%). 6 participants failed the preliminary investigation/saliva swallow, and 30 failed one of the three bolus swallow conditions. Aspiration (GUSS sum scores ≤14) was identified in 25 participants (49%) (Figure 1).

### 3.2 FEES results

All FEES examinations were tolerated without complications. Scores were allocated with the PAS with strong inter-rater reliability \((r_i = 0.866, p = 0.000)\). FEES identified impaired swallowing in 37 participants (73%) (penetration or aspiration PAS ≥3). Out of these, 14 participants (38%) aspirated at least one of the administered bolus textures (PAS ≥6). Silent aspiration was observed in 11 participants (30%).

### 3.3 Inter-rater reliability of the GUSS in patients with PD

Four GUSS videos had to be excluded from the inter-rater analysis due to insufficient acoustic quality. The two independent GUSS ratings of the remaining 47 participants showed high inter-rater reliability \((r_i = 0.8; p < 0.001)\). Disagreements resulted from different ratings on the parameters coughing (voluntary cough or cough after swallow) and voice change. Ratings of rater #1 (that had been obtained online during the examination) were used for further analyses.
The GUSS detected aspiration with a sensitivity of 50% and a specificity of 51.3% (Table 3). The accuracy of the GUSS to predict aspiration in PD was low (AUC = 0.482, SE = 0.088, p = 0.841; 95%CI = 0.309–0.655).

Dysphagia/penetration was detected with a sensitivity of 73% and specificity of 35.7% (Table 4). The accuracy of the GUSS to predict dysphagia/penetration in PD patients was low (AUC = 0.437, SE = 0.092, p = 0.493; 95%CI = 0.257–0.617).

### 3.5 Diet recommendations: GUSS vs. FEDSS vs. self-report

No significant agreement was found between the GUSS vs. FEDSS diet recommendations ($r_s = 0.12, p = 0.42$). Both scales recommended tube feeding in 50% of the cases with GUSS allocating NPO in 21 patients (41%) and FEDSS in 4 patients (8%) (Figure 2). Consistent NPO (Nil per Os) recommendation by both assessment tools was found in only 1 case, whereas 20 participants were recommended NPO by the GUSS but not by the FEDSS.
Both scales had a low agreement with the self-reported current feeding status of the participants: All patients indicated to be on an unrestricted oral diet or had moderate diet modifications without pulmonary complications within a one-year period preceding data collection.

4 | DISCUSSION

Swallowing impairments are frequent in patients with Parkinson's disease and may occur long before patients notice and report their difficulties. There is a need for disease-specific screening tools that allow for repeated evaluation and early identification of aspiration and dysphagia to ensure timely and efficient referral to instrumental assessment. In our study, the applicability of the Gugging Swallowing Screen (GUSS) in patients with PD was investigated by establishing inter-rater agreement and determining the diagnostic accuracy to detect aspiration and dysphagia when applied in this patient group.

Inter-rater reliability of the GUSS in our cohort of PD patients was good, and by this, previous results in stroke patients were confirmed.

Aspiration was identified with a sensitivity of 50% and specificity of 51% indicating that the GUSS in its current form is not applicable as a screening instrument to predict aspiration in PD patients with acceptable accuracy. Dysphagia/penetration was identified with higher sensitivity (73%); however, this is considerably lower than in stroke patients (>96%) and below our determined accuracy threshold (≥80%). Furthermore, with a specificity of 36% in detecting dysphagia/penetration, the intended benefit of a dysphagia screening tool—that is, to enhance efficiency of referral to further instrumental assessments—cannot be met, and over-referral is very likely. While likelihood ratios indicate that the GUSS is associated with the occurrence of aspiration and dysphagia in this patient group, the diagnostic contribution to establish these diagnoses is rather small. Therefore, the test should not be used as the only screening measure but in combination with expert clinical and instrumental swallowing examination.

Our data replicate previous findings that the GUSS' diet recommendations overestimate the need for oral diet restrictions. Notably, the NPO (Nil per Os) recommendation was in accordance with the FEES result in only 1 case, whereas 20 participants were recommended NPO incorrectly by the GUSS. This result should be interpreted as further emphasizing the need for instrumental and expert clinical swallowing examination in PD patients, when abnormal findings can be identified during screening procedures. Clinicians will typically make more conservative diet recommendations on the basis of a screening test result alone in comparison with those made on a gold standard test. Too restrictive diet recommendations might increase the risk of malnutrition and non-use complications particularly in individuals with progressive diseases such as PD.

Our results also provide approaches to possible modifications of the GUSS that might enhance its applicability in PD patients. The most crucial GUSS parameters were “coughing,” “voice change” and “delayed swallowing,” while “reduced vigilance,” “drooling” and “absent swallowing response” were not detected in any patient. These findings corroborate previous results. The low specificity of the GUSS may be partly due to its focus on the perceptual evaluation of “effective coughing” and “voice change” after bolus intake. This is probably a crucial discriminating factor of swallowing screening tests in stroke vs. PD patients. While the association between aspiration risk and impaired cough function is well-established, the accuracy of perceptual assessment of cough effectiveness is less clear. Acoustic evaluation of cough strength is less reliable than measuring cough characteristics such as peak cough flow (PCF), peak expiratory flow (PEF), or cough sensitivity (e.g., inhalation of citric acid or capsaicin). Inclusion of these methods into swallowing examination in...
PD patients has been widely suggested\(^\text{25-27}\) and might increase the validity of the GUSS as a dysphagia screening tool in this patient group.

“Wet voice” or “voice change” is a commonly used indicator in swallowing screening tests,\(^\text{28}\) however, the validity of this parameter to identify aspiration is not straightforward. Some studies indicate that inclusion of this parameter enhances sensitivity in water-swallowing tests,\(^\text{29}\) others found that sensitivity varies with increasing volumes and different bolus consistencies in multi-texture screening tools.\(^\text{30}\) Voice quality is particularly difficult to assess in patients with PD as their voice quality deteriorates as the disease progresses and impairments in vocal cord function can interfere with perceptual evaluation of wet voice.\(^\text{31}\) In a study with PD patients and experienced raters, wet voice was the least perceived vocal abnormality compared to hoarseness and tremor and its inclusion into a multi-texture swallowing screening contributed to specificity but not to sensitivity in detecting aspiration.\(^\text{32}\) Thus, future research should further establish the predictive properties of this parameter when included in a multi-texture screening tool for PD patients.

“Delayed swallowing” was the predominant symptom in the 26 patients reaching the GUSS’ solid bolus condition in our study, with a mean OTT (first bite of the bread until first swallow response) of 20 seconds (SD = 7.74) which confirms previously reported prolongation of OTT.\(^\text{4}\) None of our patients was able to complete the test in <10 seconds as defined in the GUSS protocol,\(^\text{11,32}\) and 10 patients could not manage the bread in ≥23 seconds (the cutoff point defined in the GUSS manual). Based on established normative data,\(^\text{33}\) a mean OTT for solid bolus material of 17.56 seconds (95% CI = 10.17–24.96) for men and 15.65 seconds for female (95% CI = 9.31–21.98) could be expected in the age group included in our study. Thus, further research is warranted to determine valid cutoff thresholds for OTT and delayed swallowing in PD patients.

4.1 | Limitations

The GUSS was developed and validated for acute stroke patients, and the same applies to the FEDSS. Thus, a direct transfer of these procedures to PD patients might be questionable in the first place. However, we aimed to investigate the applicability of an existing and widely applied, validated multi-texture screening tool in this patient group. Data were collected during a short time of patient recruitment, while a longer data collection period could have enhanced the sample size in our study. However, this limitation is partly balanced by the distribution of H&Y severity stages in our sample corresponding to the normal distribution across the population.

5 | CONCLUSION

The multi-texture screening tool GUSS in its current form, although applicable with good inter-rater reliability, is not able to detect aspiration in PD patients with sufficient accuracy. We suggest a modified version of the GUSS that incorporates adaptations of the parameters cough effectiveness, voice change, and delayed swallowing to account for the complex pathophysiology of swallowing disorders in PD patients. Our data confirm that diet recommendations drawn from screening tools should be verified by adequate clinical and instrumental assessment approaches as the complex progressive limitations of swallowing physiology in PD patients need a holistic approach to dietary adaptations.

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AUTHOR CONTRIBUTIONS

UF, JR, CP, JCN, and AN performed study conception and design, data collection, data analysis and interpretation, drafting/revising manuscript, including medical, and final approval. BS and JCK performed data collection and data interpretation and contributed to drafting and revising the manuscript. CP, CB, MPN, and CG were clinical supervisors, involved in protocol writing, experimental session, and interpretation of data and also contributed to drafting and revising the manuscript.

DISCLOSURE

The authors declare that they have no conflict of interest.

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