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Analysis of the Prevalence and Onset of Dysphonia and Dysphagia Symptoms in Movement Disorders at an Academic Medical Center

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Abstract

Voice and swallowing impairments are common in movement disorders, but their effect on patients' quality of life is not well known. This study was conducted to determine the onset and prevalence of patient-reported dysphonia and dysphagia symptoms in Parkinson's disease (PD), dystonia, Atypical Parkinsonian Syndromes (APS), and Essential Tremor (ET). Patients referred to a movement disorders clinic in a tertiary care academic medical center completed validated voice and swallowing specific Quality of Life (QOL) questionnaires: Voice Handicap Index-10 (VHI-10) and Eating Assessment Tool-10 (EAT-10). Patient demographics and clinical data were also collected. Two hundred and sixty-eight patients (males=150, females=118) completed the questionnaires (n was PD=103, APS=30, ET=56, dystonia=32, other=47). Prevalence of patient-reported dysphagia symptoms was significantly higher in APS (63%) than PD (26%), ET (25%), and dystonia (31%). Prevalence of patient-reported dysphonia symptoms was significantly lower in ET (14%) compared to PD (34%) and APS (43%). Disease duration was shorter in PD and APS compared to ET and dystonia ($p<0.05$) before reporting clinically significant dysphonia and dysphagia symptoms indicating an earlier onset of these symptoms. There were significant positive correlations between VHI-10 and EAT-10 scores and disease severity, as indicated by Unified Parkinson's Disease motor scores ($p<0.0001$) and modified Fahn-Tolosa-Marin Tremor Rating sub-scores ($p=0.0013$). Patient-reported dysphonia and dysphagia symptoms were present in one fourth of patients with PD, ET, dystonia, and almost two thirds in APS. Patient-reported QOL measures, such as VHI-10 and EAT-10, can help screen movement disorder patients for dysphonia and dysphagia symptoms.

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Keywords

Parkinson's disease; essential tremor; dystonia; atypical parkinsonian syndromes; dysphagia; dysphonia; patient-reported outcome measures

INTRODUCTION

Parkinson's disease (PD), Essential Tremor (ET), dystonia, and Atypical Parkinsonian Syndromes (APS) can interfere with the preliminary motor, behavioral, and sensory acts needed for normal voicing, speech, and/or deglutition [1–4]. Imbalance between the subsystems used in voicing, namely that of the actuator (breath management), vibrator (biomechanical function of the vocal folds), and resonator (manipulation of the shape of the vocal tract to alter voice quality) can negatively impact vocal quality, consistency, pitch, intensity, or ease of phonation [5–6]. Dysphonia is when vocal quality does not match the expected norms for the patient's age, gender, culture, or when there is change from what the patient defines as their "normal" voice [5–6]. The act of swallowing is divided into stages: the oral preparatory phase, oral phase, pharyngeal phase, and esophageal phase [7]. Discoordination of muscles activated in any of these phases can result in dysphagia, or difficulty moving food or liquid from the mouth to the stomach [7]. Dysphagia can lead to poor nutrition or aspiration, which is the entry of material between the vocal folds into the lower airway; in some cases, it can lead to pneumonia or morbidity [7].

Due to the difference in disease progression and pathology that exist between PD, ET, dystonia, and APS, each of these diseases have unique clinical profiles for the onset, severity, and symptomology of dysphagia and dysphonia [8–9]. Normal voice function is one of the first components to begin deteriorating in PD [10–11] with articulation and fluency impairment reported in 50–85% of the patients and 40–75% of PD patients experience swallowing dysfunction [12–14]. Patients with ET tend to experience perturbations in vocal intensity and frequency that produce a voice tremor [15]. Patients diagnosed with APS, such as patients with Multiple System Atrophy (MSA) and Progressive Supranuclear Palsy (PSP), more frequently experience speech, swallowing, and voice dysfunction than patients with PD [4].

The patient-reported quality of life impact of dysphonia and dysphagia in PD, APS, ET and dystonia has not been well studied. Patient-reported outcome measures offer some important advantages over clinician assessments – they focus on how the patient feels their symptoms and disease are impacting their life while avoiding rater-based bias [16–19]. We used the Voice Handicap Index-10 (VHI-10) and Eating Assessment Tool-10 (EAT-10) which are validated Quality of Life (QOL) surveys for patients to report the subjective presence and degree of dysphonia [18] and dysphagia [19]. We combined these scales with clinician-administered assessments to analyze differences in dysphonia and dysphagia severity, onset, and prevalence between movement disorders (APS, ET, PD, dystonia). To our knowledge, this is the first study to utilize these measures for patient-reported outcome measures in a movement disorder population.

METHODS

Subjects

Completed QOL surveys from 268 consecutive movement disorder patients (males=150, females=118) were obtained from three fellowship-trained movement disorder specialists in the Department of Neurology at Wake Forest Baptist Medical Center. A retrospective chart review was completed to obtain diagnosis, demographics, physician-administered disease severity scales, QOL scores, and disease duration. The inclusion criteria were: 1) Patients diagnosed with PD, ET, dystonia, or APS by a fellowship-trained movement disorder neurologist; 2) completion of the QOL survey consisting of the VHI-10 and EAT-10; 3) age above 18 years. The 'Other' group, consisting of movement disorders other than PD, ET, dystonia and APS, was excluded from the analysis due to small numbers in each disease category.

Study Design

This study was approved by the Institution Review Board (IRB00039291). Patients were assigned to one group based on the primary disease diagnosis they received from a fellowship-trained movement disorder neurologist. The clinical characteristics of patients who met the inclusion criteria are described in Table 1. The dystonia category consists of patients diagnosed with Meige's syndrome (n=1), focal dystonia (n=2), multifocal dystonia (n=1), generalized dystonia due to DYT-1 mutation (n=1), generalized dystonia without mention of a specific genetic cause (n=7), and cervical dystonia (n=20). The Atypical Parkinsonism category consists of patients diagnosed with Vascular Parkinsonism (n=8), Multiple System Atrophy (n=5), Corticobasal Degeneration (n=4), Progressive Supranuclear Palsy (n=3), drug-induced Parkinsonism (n=5), and Lewy Body Degeneration (n=5). The Other category consists of patients diagnosed with functional movement disorders (n=14), myoclonus (n=8), hemifacial spasm (n=1), ataxia (n=19), orthostatic tremor (n=2), tardive dyskinesia (n=2), chorea (n=1).

Assessment

Patients completed the VHI-10 [18] and EAT-10 [19]. The VHI-10 is a validated 10-question instrument for patient self-assessment of dysphonia symptoms that encompasses multiple aspects of voice dysfunction, including functional and emotional aspects [18]. The EAT-10 is a validated 10 question self-assessment instrument designed to assess the patient's perception of their dysphagia severity [19]. Clinician-administered Unified Parkinson's Disease Rating Scale (UPDRS) motor scores [20] were collected for PD and APS. Clinician-administered modified Fahn-Tolosa-Marin Tremor Rating Scale (mTRS) [21] and Burke-Fahn-Marsden Dystonia Rating Scale – Movement/Disability (BFMDRS-M/D) [22] scores were collected for ET and dystonia respectively. Patients with PD were further analyzed based on the stage of the disease by using the Modified Hoehn-Yahr scale [23].

Scoring

A score of 12 or greater on the VHI-10 indicates clinically significant dysphonia [18] and a score of 10 or greater indicates that a referral to a Speech Language Pathologist may be

needed for further evaluation [18]. A score of 4 or greater on the EAT-10 is considered abnormal and indicative of clinically significant dysphagia [19]. However, a score on either the VHI-10 or EAT-10 cannot be directly translated to degree of dysphagia, voice impairment, or tremor. Clinician-administered scales were also utilized to assess this movement disorder population. The specific speech components of the UPDRS, mTRS, and BFMDRS-M/D allows the physician to objectively grade the patient's speech from 0–4 with 0 being normal speech and 4 being unintelligible most of the time.

Statistical Analysis

The statistical software used for all statistical analysis was SAS version 9.4. Between-diagnoses differences in patient demographic characteristics were assessed using Chi-square tests for categorical variables and analysis of variance (ANOVA) for continuous variables. Significant omnibus differences were followed up with pairwise comparisons for age and disease duration to determine which disease groups were significantly different and 95% confidence intervals were calculated. For patients above the clinically significant values for the VHI-10 and EAT-10, a one-way ANOVA was used to compare the diagnostic groups on their disease. Correlations were used to assess the relationship between disease duration, EAT-10 and VHI-10 scale scores, and UPDRS and mTRS scales. Due to a small number of individuals who were on medications, it was only possible to analyze off-medication scores for the mTRS. A chi-squared goodness of fit test was used to determine the differences in dysphonia and dysphagia symptoms between PD, APS, ET, and dystonia.

RESULTS

Patient characteristics are shown in Table 1. Patients with PD were also analyzed based on their stage in the Modified Hoehn-Yahr scale. Out of the total 103 patients with PD, 1 patient was in Stage 1 (0.97%), 15 patients were in Stage 1.5 (14.56%), 26 patients were in Stage 2 (25.24%), 21 patients were in Stage 2.5 (20.39%), 35 patients were in Stage 3 (33.98%), 2 patients were in Stage 4 (1.94%), and 3 patients were in Stage 5 (2.91%). The majority of patients (94.17%) were neither in late (Stage 4–5) nor early stages (Stage 1) of PD.

Comparison of Disease Duration with VHI-10 and EAT-10 Clinically Significant Scores

We compared average disease duration (years) for the patients in each disease group who reported clinically relevant symptoms of dysphonia and dysphagia, defined as a VHI-10 scale score ≥ 12 and/or EAT-10 scale score ≥ 4 (Figure 1). Table 2 contains the descriptive statistics regarding the mean disease duration for VHI-10 and EAT-10 scales for each disease group.

Disease diagnosis had an effect on both the VHI-10 ($F_{4, 75} = 3.6, p = .009$) and EAT-10 ($F_{4, 86} = 4.5, p = .002$) scores. Patients with ET had significantly longer disease durations before reporting clinically significant VHI-10 and EAT-10 scores when compared to PD and APS (Table 3). Patients with dystonia also had significantly longer disease durations before reporting clinically relevant VHI-10 scores compared to APS, as well as EAT-10 scores compared to patients with PD and APS (Table 3). When comparing patients with PD and

APS, there were not significant differences in average disease duration before reporting clinically relevant VHI-10 or EAT-10 scores.

Correlational Analysis

VHI-10 and EAT-10 Scores Correlated with Disease Duration in Patients with PD: PD patients with a longer disease duration tended to have significantly higher VHI-10 ($r = 0.4$, $p = 0.0005$, $n = 73$) and EAT-10 scores ($r = 0.42$, $p = 0.0002$, $n = 73$). A correlation between disease duration and VHI-10 and EAT-10 scores was not observed in any of the other disease groups.

UPDRS Correlation with VHI-10 and EAT-10 Scores: UPDRS motor scores and the speech subsection of the UPDRS scale (both on and off PD medications) were compared with VHI-10 and EAT-10 scores. Off-medication motor UPDRS scores had a significant, positive correlation with severity of VHI-10 ($r = 0.45$, $p < .0001$, $n = 137$) and EAT-10 scores ($r = 0.33$, $p < .0001$, $n = 137$); off-medication speech sub-scores of the motor UPDRS scale had a significant, positive relationship with VHI-10 ($r = 0.52$, $p < .0001$, $n = 137$) and EAT-10 scores ($r = 0.28$, $p = 0.0008$, $n = 137$). For on-medication results, only the speech sub-scores of the motor UPDRS scale had a significant, positive relationship with VHI-10 ($r = 0.50$, $p = 0.0001$, $n = 54$) and EAT-10 scores ($r = 0.31$, $p = 0.024$, $n = 53$).

mTRS Correlation with VHI-10 and EAT-10 Scores: mTRS scores (off tremor medications only) were compared with VHI-10 and EAT-10 scores. Off-medication head tremor sub-scores of mTRS had a positive correlation with severity of EAT-10 ($r = 0.28$, $p = 0.054$, $n = 49$) scores and VHI-10 scores ($r = 0.19$, $p = 0.18$, $n = 49$), although not significant. Off-medication voice tremor sub-score of mTRS scores had a significant, positive correlation with VHI-10 scores ($r = 0.45$, $p = 0.0013$, $n = 48$), as well as a positive correlation with EAT-10 scores ($r = 0.26$, $p = 0.072$, $n = 48$).

Frequency of patient-reported Dysphonia Symptoms Between Disease Types

Patient-reported prevalence of dysphonia was 34% in PD, 43% in APS, 14% in ET, and 28% in dystonia (Figure 2). ET had a lower prevalence of patient-reported dysphonia symptoms when compared to PD and APS ($p = 0.0076$, $p = 0.0028$, respectively).

Frequency of patient-reported Dysphagia Symptoms Between Disease Types

Patient-reported prevalence was 26% in PD, 63% APS, 25% in ET, and 31% in dystonia (Figure 2). APS had a higher prevalence of patient-reported dysphagia symptoms than PD, ET, and dystonia ($p = 0.0002$, $p = 0.0006$, $p = 0.0114$, respectively).

DISCUSSION

It is crucial to accurately measure speech, voice, and swallowing function in movement disorder populations. When these processes fail to function normally, they are debilitating [24]. Although clinician-administered scales allow the physician to assess physician-perceived dysarthria, dysphagia, and dysphonia, patient-reported measures allow a truer reflection of how the patient feels they are impacted by their debility. Combining patient-

reported measure with clinician-administered assessments provides a more exhaustive and complete view into a patient's health. Patient-reported QOL measures allow us to directly examine patient perceptions of impairment and dysfunction resulting from dysphonia and dysphagia symptoms, making them essential for one of the primary goals of patient care – helping patients feel better.

In this study, we observed a significant difference in the prevalence of patient-perceived symptoms of dysphonia and dysphagia, with ET displaying significantly lower rates of patient-perceived dysphonia symptoms and APS displaying significantly higher patient-reported dysphagia symptoms. These results are consistent with the literature showing that patients with ET generally experience voice tremor, but not severe speech and voice symptomology [2, 25]. Patients with PD and APS reported clinically significant dysphonia and dysphagia symptoms at earlier disease durations than ET and dystonia. This is also expected given the faster progression of symptoms in APS [4, 26] and that speech and voice functions become impaired in PD [10–11]. While we did find that a higher percentage of patients with APS reported clinically significant VHI-10 and EAT-10 scores compared to PD, and that patients with APS also reported them sooner than patients with PD, we did not find a significant difference between these groups in the average disease duration before reporting clinically significant scores. This result could be contributed to the heterogeneity of patients within the APS disease group in this study.

The VHI-10 and EAT-10 scales are frequently utilized in otolaryngology literature for various uses, such as determining treatment effects [27] or developing sub-scales to assess patient-perceived symptoms in specific subpopulations like the Signing Voice Handicap Index [28]. However, neurology literature primarily focuses on speech dysfunction by examining dysarthria [10, 12–13], which is obtained through clinician assessment via the BFMDRS, the UPDRS Motor and/or Activities of Daily Living, or the mTRS. Kalf et al [29] conducted a meta-analysis on the prevalence of oropharyngeal dysphagia in patients with PD. From their literature review of 350 articles, they found 10 which utilized either a patient-completed survey or interview for dysphagia. These studies utilized scales do not give a complete picture of patient voice and swallowing dysfunction as the surveys or interview questions utilized only contain 1–2 questions, in which the subjects can answer either yes or no (PD NMSQuest), or never, sometimes, regularly, or often (SCOPA-AUT); dysphonia was not assessed. Frequency and classification of dysarthria [10–13] is commonly discussed in Parkinson's disease, but dysphonia is rarely examined. Dysphonia is discussed in ET literature, but mostly in the context of response to botulinum toxin [30]. One study examined 34 patients with ET to characterize vocal tremor. They utilized the Vocal Tremor Scoring System as well as the VHI scale and reported 23 patients having vocal tremor, and that this symptom typically did not appear until patients were 70 years old [2]. Dysphagia is also explored in this patient population with results showing that patients with ET frequently do not suffer from debilitating dysphagia [31].

Dysphonia is discussed in dystonia literature, but primarily in terms of the focal dystonia adductor or abductor spasmodic dysphonia [32]; one study focusing on dysphagia utilized electrophysiological evaluation to report 72% of patients with dystonia displayed pharyngeal dysphagia [33]. Lastly, early onset of dysphagia is discussed in APS literature as a method to

help distinguish APS from PD with most studies reporting deterioration of swallowing function in as early as the first year of APS onset [4, 26]. Current literature on these disease populations does not adequately compare differences in the onset, characteristics, and prevalence of dysphonia and dysphagia by relying on patient-reported outcome measures to assess how patients feel they are impacted by their voice and swallowing dysfunction. Our study is novel, owing to our utilization of both subjective patient-reported outcome measures and clinician-administered scales to compare differences in onset and frequency of dysphagia and dysphonia symptoms between four distinct movement disorder categories.

Identification of dysphagia with patient-reported surveys can help in prompt referral for swallowing evaluation and prevention of aspiration pneumonia [34]. Our study highlights that many patients with PD are not aware of this issue, as is indicated by the low percentage (26%) which implies a deep need for education, specifically because this patient population frequently succumbs to aspirational pneumonia [34]. Our study not only reports that dysphagia symptoms are common, but also provides insight into when clinicians and patients can begin experiencing these symptoms based on the disease duration. These values can be used to help educate patients, especially patients with APS, who had the lowest average disease duration for VHI-10 (4.30 years) and EAT-10 (5.89 years), indicating that these patients are more likely to experience symptoms of dysphonia and dysphagia earlier than other disease groups in this study.

Our study is limited by its retrospective design. Only patients who fully completed the survey were included, though we addressed this limitation with large sample sizes for PD, ET, APS and dystonia. Moreover, utilizing a patient-completed survey introduces the possibility of subject not providing fully complete and/or accurate answers; however, this limitation is also addressed by our large sample size of 268 subjects. Encouraged by our results, we are studying effects of Deep Brain Stimulation (DBS) on patient-perceived symptoms of dysphonia and dysphagia with the help of the VHI-10 and EAT-10.

CONCLUSION

To our knowledge, no prior study has utilized specific dysphonia and dysphagia patient perception scales in a cohort of patients with PD, APS, ET, and dystonia to determine the prevalence and onset of patient-perceived dysfunction. Patients with PD and APS both had significantly higher patient-reported dysphonia symptoms compared to patients with ET. Patients with APS had significantly higher patient-reported dysphagia symptoms and shorter disease duration before reporting these symptoms than patients with PD, ET, and dystonia. This study exhibits the importance of utilizing patient completed QOL surveys to gather information about symptoms that may be secondary to a movement disorder yet can require medical intervention.

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- Patients with Parkinson's Disease show a low awareness of dysphagia symptoms.
- Essential Tremor patients report a lower prevalence of dysphonia symptoms.
- Atypical Parkinsonian Syndromes report a higher prevalence of dysphagia symptoms.
- Atypical Parkinsonian Syndromes report dysphagia and dysphonia symptoms earliest
- Correlation between dysphagia and dysphonia scores with disease severity scores.

Figure 1. Comparison of Average Disease Duration (in years) for Patients Achieving Clinically Relevant Scores for VHI-10 and EAT-10. Patients with ET and dystonia had significantly longer disease durations before reporting clinically significant VHI-10 and/or EAT-10 scores when compared to PD and APS (see Table 2 for mean difference with 95% confidence interval).

Figure 2.

Frequency of patient-reported dysphonia and dysphagia symptoms between patients with PD, ET, APS, and dystonia: ET had lower prevalence of patient-reported dysphonia symptoms when compared to PD and APS ($p = 0.0076$, $p = 0.0028$, respectively). APS had a higher prevalence of patient-reported dysphagia symptoms when compared to PD, ET, and dystonia ($p = 0.0002$, $p = 0.0006$, $p = 0.0114$, respectively).

Table 1.

Patient characteristics of movement disorder patients that completed the VHI-10 and EAT-10 surveys and met the inclusion criteria for this study.

Primary Movement Disorder Diagnosis	Number of Subjects	Average Age $\pm$ standard deviation	Average Disease Duration $\pm$ standard deviation
Parkinson's disease	103 (males=67%, females=33%)	68.17 $\pm$ 9.93	5.92 $\pm$ 4.46
Atypical Parkinsonian Syndromes	30 (males=63%, females=37%)	70.63 $\pm$ 8.91	5.37 $\pm$ 7.20
Essential Tremor	56 (males=54%, females=46%)	68.63 $\pm$ 9.09	20.23 $\pm$ 16.59
Dystonia	32 (males=28%, females=72%)	62 $\pm$ 9.98	12.42 $\pm$ 11.02
Other	47 (males=49%, females=51%)	56.04 $\pm$ 13.66	7.50 $\pm$ 9.84

Table 2.

Average disease duration for PD, ET, Dystonia, and APS patients who reported clinically relevant VHI-10 and EAT-10 scores.

Disease Diagnosis	Participants reporting clinically relevant VHI-10 scores	VHI-10 average disease duration $\pm$ standard deviation	Participants reporting clinically relevant EAT-10 scores	EAT-10 average disease duration $\pm$ standard deviation
PD (N=103)	N=35 (34%)	7.43 $\pm$ 3.85	N=27 (26%)	7.07 $\pm$ 3.45
ET (N=56)	N=8 (14%)	16 $\pm$ 11.16	N=14 (25%)	17.86 $\pm$ 13.59
Dystonia (N=32)	N=9 (28%)	11.88 $\pm$ 9.69	N=10 (31%)	15.90 $\pm$ 14.91
APS (N=30)	N=13 (43%)	4.30 $\pm$ 3.09	N=19 (63%)	5.89 $\pm$ 8.58

Table 3.

Disease duration before reporting significant dysphagia and dysphonia symptoms. Shown are the differences in disease duration for the disease groups showing a significant ($p < 0.05$) difference in the average disease duration (in years) before reporting clinically relevant scores in the VHI-10 and EAT-10.

VHI-10				
Disease Comparison	ET - PD	ET - APS	Dystonia - APS	
Difference in Average Disease Duration [95% CI]	8.57 [2.65, 14.50]	11.69 [4.90, 18.50]	7.58 [1.03, 14.14]	
EAT-10				
Disease Comparison	ET - PD	ET - APS	Dystonia - APS	Dystonia - PD
Difference in Average Disease Duration [95% CI]	10.78 [4.22, 17.35]	11.96 [4.94, 18.98]	10.01 [2.23, 17.79]	8.83 [1.45, 16.21]