# SELF-PERCEIVED AFFECTIVE, BEHAVIORAL, AND COGNITIVE REACTIONS ASSOCIATED WITH VOICE AND COMMUNICATION IN PEOPLE WITH PARKINSON'S DISEASE

By

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#### **APPROVAL PAGE**

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#### I. Introduction

#### Literature Review, and Significance

#### Voice Disorders and Voice Therapy Outcomes in Different Settings

Voice disorders impact the phonatory or respiratory system and lead to voice quality deviating from what would be expected in an individual without dysphonia of the same age, gender, and culture (Aronson & Bless, 2011; Ramig & Verdolini, 1998). Voice disorders are a common problem within the general population: 30% of the population will present with a voice disorder at some point in their lives (Roy et al., 2005). Within the United States, the prevalence of voice disorders has been estimated at 7% for the adult population (Bhattacharyya, 2014; Roy et al., 2005), which can be higher in populations of professional voice users (Lerner et al., 2013; Pestana et al., 2017; Phyland & Miles, 2019). Voice disorders can negatively impact the quality of life, social participation, and work performance of affected individuals (Cohen, 2010; Ramig & Verdolini, 1998). Voice disorders also are a significant cost to individuals and society. Cohen et al. (2012a) estimated that the direct costs per person per year for diagnosing and managing laryngeal disorders lie between \$577.18 and \$953.21, which amount to over 11 billion dollars annually across the United States (Cohen et al., 2012a). Given the prevalence and cost of voice disorders, their treatment via rehabilitation is important to consider. Behavioral voice therapy administered by speech-language pathologists (SLPs) is often the first line of treatment for voice disorders and is often incorporated as adjuvant to medical or surgical treatment (Desjardins et al., 2017; Ramig & Verdolini, 1998).

Speech-language pathologists with specialized knowledge and skills in voice and upper airway disorders who work primarily with a voice disordered population are referred to as "vocologists." These specialized SLPs can work in a variety of settings, such as, but not limited to, clinics led by laryngologists associated with academic medical centers or private practice community voice clinics (Cohen et al., 2012b; LeBorgne & Donahue, 2019; Watts & Knickerbocker, 2019). The disorders treated in the specialty voice clinics associated with medical centers are similar to those in a private practice voice clinic and include, among others, mid-membranous lesions, vocal fold immobility, and functional voice disorders such as muscle tension dysphonia (Misono et al., 2016; A. Remacle et al., 2017; Van Houtte et al., 2010; Watts & Knickerbocker, 2019). These epidemiological patterns within different clinics are important for several reasons: they can indicate differences in the incidence and prevalence of vocal disorders in certain populations and thus inform clinicians on the knowledge and skills needed in these settings (Watts & Knickerbocker, 2019).

Regardless of the setting or disorder type, voice therapy aims to improve vocal function so the patient's voice meets their occupational, emotional, and social needs (Aronson & Bless, 2011). This translates into treatment objectives and discharge criteria associated with the patient's ability to use improved voice quality independently and to function with the new voice in daily life (Gillespie & Gartner-Schmidt, 2018). In clinical practice, therapists use treatment plans with multiple direct and indirect voice rehabilitation techniques individualized to each client to achieve these treatment goals (Burg et al., 2015; Chan et al., 2013). However, the available literature on the effectiveness of voice therapy does not reflect this clinical reality. For example, existing randomized controlled trials were conducted in highly controlled environments that often focused on one therapeutic technique in a specific population (e.g., one type of etiology) (Carding et al., 2017; Desjardins et al., 2017). In these settings, voice therapy has been shown to lead to improvements in perceptual

voice measures, acoustic voice measures, and self-assessments of vocal parameters (e.g., vocal quality of life) (Desjardins et al., 2017). However, as these trials do not reflect realistic voice therapy practice, the extent to which these results are replicated in settings with less controlled populations and are translatable therapy approaches, such as in a private practice community voice clinic, is still unclear.

#### Voice Disorders in Parkinson's Disease

One specific population of interest often treated in voice clinics are people with Parkinson's disease (PWPD). Parkinson's disease (PD) is the most common neurodegenerative disease after Alzheimer's disease and has an increasing prevalence (Sveinbjornsdottir, 2016). In 2018, Marras et al. (2018) estimated that about 930,000 people in North America will be affected by PD in 2020. PD is a chronic and progressive neurodegenerative disease caused by the formation of Lewy bodies ( $\alpha$ -synuclein deposits) within the peripheral and central nervous systems and the subsequent degeneration of the central nigrostriatal pathways (Braak et al., 2003; Poewe et al., 2017; Sveinbjornsdottir, 2016). The neuropathology results in both motor and non-motor symptoms (Sveinbjornsdottir, 2016). The hallmark motor symptoms in PD are rest tremor (i.e., an oscillation in the muscles at 4-6Hz), rigidity (i.e., increased resistance to movement), bradykinesia (i.e., slowness of movement), and postural instability (Bartels & Leenders, 2009; Jankovic, 2008; Sveinbjornsdottir, 2016). The motor symptoms typically emerge unilaterally in limb musculature and progress to a bilateral presentation over time (Bartels & Leenders, 2009; Sveinbjornsdottir, 2016). The non-motor symptoms, on the other hand, include autonomic dysfunctions, sleep disturbances, neuropsychiatric problems, and sensory issues (Jankovic, 2008; Sveinbjornsdottir, 2016). Concomitant with the motor and non-motor symptoms, approximately 90% of the PWPD manifest speech impairments during the disease process (Ramig et al., 2004; Schalling et al., 2017).

The speech disturbances associated with PD are collectively referred to as hypokinetic dysarthria (Ramig et al., 2004; Sveinbjornsdottir, 2016). Hypokinetic dysarthria can perceptually be described as having a harsh and breathy voice with reduced intensity, having imprecise articulation, and having disturbances in speech rate and prosody (Brabenec et al., 2017; Magee et al., 2019; Ramig et al., 2004). Consequently, PD affects all subsystems of speech. Specifically for the phonatory system, hypophonia is the distinctive feature of Parkinsonian speech (Miller, 2017). Hypophonia encompasses perceptually salient lowering of vocal loudness, changes in vocal pitch, monotony of pitch and loudness, as well as harsh and breathy voice quality (Holmes et al., 2000; Magee et al., 2019). A vocal tremor can be present as well (Miller, 2017; Sewall et al., 2006). These perceptual changes are also discernable acoustically as decreased intensity, reduced fundamental frequency variability, compressed intensity ranges, lower harmonics-to-noise ratio, and increased phonatory perturbation (Gamboa et al., 1997; Holmes et al., 2000). When looking at the vocal folds using videostroboscopy, glottal insufficiency, bowing, decreased vibratory amplitude, or abnormalities of the mucosal wave can be observed (Bauer et al., 2011; Blumin et al., 2004; Merati et al., 2005; Sewall et al., 2006; Smith et al., 1995).

Multiple theories have emerged to explain the voice changes that occur in PD. Recently, these theories have focused on the role of central processing issues arising from basal ganglia dysfunction, though not necessarily dopamine deficiency (Miller, 2017; Sapir, 2014), which is the hallmark pathology in PD (Sveinbjornsdottir, 2016). The central processing issues in PWPD inhibit control of vocal output: PWPD present with underscaling

of movements and decay of vocal effort as well as difficulty maintaining adequate vocal effort for voicing resulting in a low-intensity voice signal (Miller, 2017; Sapir, 2014). Impairments to initiation and preprogramming of speech (resulting in hesitancy or difficulty starting a word) and temporal processing and rhythmicity (resulting in short rushes of speech or inappropriate pauses) are also characteristic of speech in PWPD (Miller, 2017; Sapir, 2014). Speech production deteriorates when an automatic mode of control is needed (Sapir, 2014). Moreover, PWPD have issues with sensing and adapting their movements effectively for voice and speech. The internal cueing process (e.g., self-monitoring, a.k.a. "vocal vigilance," to maintain a loud voice) is impaired, while external cueing (e.g., another person or device to provide a prompt) can serve to maintain appropriate voice quality (Sapir, 2014). PWPD do not accurately perceive their voices, and vocal intensity appears especially hard for them to monitor (Miller, 2017; Sapir, 2014). Additionally, because their vocal vigilance is reduced, PWPD have difficulty correcting their errors (Sapir, 2014). The above-described processes are likely intertwined with or influenced by each other and are not completely understood (Sapir, 2014). Sapir (2014) hypothesizes that these impaired processes are likely the predominant factor in voice changes at the beginning of the disease process when the dopamine deficiency is not yet salient. In later stages of the disease, the dopamine deficiency likely plays a predominant role in communication changes (Sapir, 2014). Dopamine deficiency can lead to the typical motor symptoms of rigidity, bradykinesia, akinesia, and tremor within the laryngeal musculature. These motor changes are hypothesized to lead to reduced vocal fold approximation and thus reduced glottal closure (A. Ma et al., 2020; Miller, 2017). The rigidity can also present as stiffer vocal folds, resulting in increases in fundamental frequency, incomplete vocal fold closure, and decreases in voice quality

(Goberman & Coelho, 2002; A. Ma et al., 2020; Zhang et al., 2005). Despite these findings, the exact mechanism that causes hypophonia remains unclear.

Interestingly, the hypophonia in PWPD changes throughout the disease process. In the late stages of the disease, the voice difficulty presents as the salient hypophonia described above: low intensity with harsh voice quality (Magee et al., 2019; Miller, 2017). However, in the early stages of PD, the hypophonia may be less perceptually salient. Acoustic voice analyses have detected greater voice irregularities in early-stage PWPD compared to healthy controls (Defazio et al., 2016; Lirani-Silva et al., 2015; Rusz, Cmejla, Ruzickova, & Ruzicka, 2011). For example, increased fundamental frequency, decreases in intensity and frequency ranges, increases in phonatory perturbation, and decreases in harmonics-to-noise ratios were apparent (Holmes et al., 2000; Lirani-Silva et al., 2015; Rusz, Cmejla, Ruzickova, & Ruzicka, 2011). Almost 80% of untreated PWPD in the early stages of the disease present with some kind of vocal impairment (Rusz, Cmejla, Ruzickova, & Ruzicka, 2011). Some changes are also likely present in the prodromal stage of the disease (Harel, Cannizzaro, & Snyder, 2004; Harel, Cannizzaro, Cohen, et al., 2004). Changes to the acoustic signal in the early stages of PD are not necessarily perceivable. For example, Lirani-Silva et al. (2015) did not find statistically significant differences between the PD group and a group of healthy controls for auditory-perceptual measures of phonation. On the other hand, Holmes et al. (2000) did find statistically significant perceptual differences between the voices of healthy controls and early-stage PWPD. Stewart et al. (1995) found increased perceived roughness, reduced loudness, as well as breathiness and monopitch in early-stage PWPD, but did not compare with controls without PD. More research is needed on how voices in non-advanced PD can be described perceptually, as auditory-perceptual assessment of vocal function is a

primary component of the clinical voice evaluation. Charting these early voice changes is important, as Gibbins et al. (2017) suggested that voice therapy may be most beneficial in the early stages of PD, as there is no compensatory behavior or obvious laryngeal pathology present yet. Moreover, longer disease duration at the inception of voice therapy has been suggested to lead to smaller gains post-treatment (Boutsen et al., 2018). However, only a few studies are available on the treatment of early-stage voice disorders, possibly because voice impairments in PWPD are not necessarily diagnosed at the beginning of the disease process due to the aforementioned issues with the saliency of the early vocal changes (Ciucci et al., 2013).

#### Psychosocial Consequences of Voice Disorders in Parkinson's Disease

The communication changes in PD can, over time, severely impact the intelligibility of speech and consequently the social participation and the quality of life in PWPD (Dashtipour et al., 2018; Gillivan-Murphy et al., 2019; Miller et al., 2008; Miller, 2017; Ramig et al., 2004). Recently, attention has been given to these psychosocial consequences of hypokinetic dysarthria. This psychosocial impact can be defined as the social and psychological factors that influence a person's quality of life, participation, and overall wellbeing (Walshe, 2010). PWPD have reported a significant psychosocial impact of PD on communication, even when the hypokinetic dysarthria is not yet perceptually salient. This perceived impact is independent of disease status, cognitive status, and intelligibility (Gillivan-Murphy et al., 2019; Miller et al., 2008; Miller et al., 2011). More specifically, PWPD can experience frustration due to communication partners misunderstanding the verbal and non-verbal messages they try to communicate (Gillivan-Murphy et al., 2019; Whitehead, 2010; Yorkston et al., 2017) as well as embarrassment because of their speech difficulties (Schalling et al., 2017; Yorkston et al., 2017). Gillivan-Murphy et al. (2019b) also emphasized that the communication changes of PD can affect the confidence of PWPD, as speech and voice reflect identity, personality, and emotional state. PWPD feel less competent, less adequate, and less in control (Gillivan-Murphy et al., 2019b; Johansson et al., 2020; Miller et al., 2011). Furthermore, PWPD need increasing physiological energy to participate in communication exchanges or may feel too fatigued to participate in communication (Gillivan-Murphy et al., 2019b; Johansson et al., 2020; Whitehead, 2010; Yorkston et al., 2017). Collectively, the communication changes throughout the disease process negatively impact PWPD's participation in work life, family life, social life, and leisure activities, leading to social withdrawal (Miller et al., 2006; Schalling et al., 2017; Whitehead, 2010).

To compensate for the negative feelings and thoughts about communication, PWPD adjust their communication behaviors. Whitehead (2010) found that PWPD actively look for coping strategies when participating in communication with others. For example, PWPD might alter physical effort to create greater vocal intensity (Miller et al., 2006). Another compensation is managing their conversational style. This could encompass waiting to speak, preparing what they want to say, talking as little as possible, or even avoiding talking and conversational strategy, PWPD can inform their environment of their speech difficulties (Miller et al., 2006; Schalling et al., 2017; Whitehead, 2010). As an additional conversational strategy, PWPD can inform their environment of their speech difficulties (Miller et al., 2006; Schalling et al., 2017). While studies have demonstrated these overt and covert compensation strategies of PWPD when communicating with others, we know very little about how PWPD are affectively and cognitively impacted or about the full extent of their coping strategies when the disease impairs their communication.

Several instruments have been used in PWPD to look at the psychosocial consequences of their communication deficits (Baylor et al., 2013; Donovan et al., 2008; Walshe et al., 2009). For example, Letanneux et al. (2013) and Cardoso et al. (2018) have used Walshe et al.'s Dysarthria Impact Profile (2009) within the PD population and found it to be a potentially useful tool. The Dysarthria Impact Profile is a valid and reliable measure with 48 questions, which aims to quantify the impact of acquired dysarthrias. It focuses on the psychological effect of dysarthria on the speaker, acceptance of dysarthria, the perception of other's reactions, and how the dysarthria impacts communication (Walshe et al., 2009). Another assessment, the Communicative Effectiveness Survey (Donovan et al., 2008; Hustad, 1999), contains eight questions focusing solely on the effectiveness of communication (Donovan et al., 2008) and has been validated for use in PWPD (Donovan et al., 2008; Dykstra et al., 2015). A third tool is the Communication Participation Item Bank (Baylor et al., 2009; Yorkston et al., 2008), which describes participation in speaking situations and has been calibrated specifically for the PD population (Baylor et al., 2013). This instrument found that the speech difficulties in PWPD significantly impacted their participation in social events (Baylor et al., 2013; McAuliffe et al., 2017), which is valuable information that compliments the standard tools for describing the communication impairment profile of PWPD (McAuliffe et al., 2017). However, all of the discussed assessments only look at specific subparts of the psychosocial wellbeing related to communication in PWPD. Specifically with regard to the Communicative Effectiveness Survey (Donovan et al., 2008; Hustad, 1999) and the Communication Participation Item Bank (Baylor et al., 2009; Yorkston et al., 2008), this means that they only consider the effectiveness of communication and the interference in communication respectively. These

assessments do not take into account the emotional or cognitive processes underlying communication exchanges. The Dysarthria Impact Profile (Walshe et al., 2009) is a more extensive instrument but describes only limited speech situations and coping behaviors. None of these described tools have norms available for healthy and disordered populations. Moreover, there is no standard tool to assess the psychosocial consequences of communication in PWPD currently used in research or clinical environments.

Typically, treatment for voice-related impairments of PWPD is unidimensional and thus focused solely on perceptual voice disturbances (Gillivan-Murphy et al., 2019b). Framed within the International Classification of Functioning, Disability, and Health (ICF, World Health Organization, 2002), the typical voice treatment in PWPD can be said to focus only on "body functions" and "activity," ignoring other important elements of this model: "participation," "environmental factors," and "personal factors" (World Health Organization, 2002). Even after therapy, frustration, anxiety, and negative attitudes can persist, as speech and voice do not completely return to normal (Spurgeon et al., 2015). Yorkston et al. (2017) have reported that PWPD want the psychosocial effects of their communication disorder included in their treatment. A comprehensive and multidimensional approach to treating voice impairments in PWPD would include the SLP assessing the cognitive aspects of the communication issues (e.g., how they think about and react to speaking situations). For example, it could consider how the individual's communication issues affect relationships and social withdrawal (Yorkston et al., 2017). The above literature review highlights the importance of incorporating psychosocial aspects of communication difficulty in PWPD during therapy. Reporting on and including these psychosocial aspects in therapy may allow

for more multidimensional and holistic therapy approaches, which also target how a speaker with PD feels and thinks when voice production is necessary.

Oral communication requires an active partner engaged in the communicative process. Communication partners close to PWPD consequently play an important role in their social life and experience the PWPD's communication difficulties first-hand (Gillivan-Murphy et al., 2019b). Additionally, PD pathology influences several neuropsychological processes that can affect self-perception (Parveen & Goberman, 2017). The PWPD's perception, consequently, may not be in accordance with those of their communication partners (i.e., communication proxy). The perceptions of communication proxies are important to understand as PWPD may have increasing difficulties when communicating, especially in the later stages of the disease. Close communication partners may need to rely on perceptions of communication difficulties to better repair communication exchanges and to improve the communication experiences of PWPD. Whitehead (2010) considered the opinion of the PWPD's spouse on the psychosocial consequences of the communication difficulties. She found that spouses are aware of the communication changes in their partners with PD (Whitehead, 2010). Partners also noticed the dysarthria's consequences such as less confidence and social withdrawal in the PWPD (Whitehead, 2010). Dykstra et al. (2015) found no difference between the ratings of the communicative effectiveness of PWPD themselves and their primary communication partners. Miller et al. (2008), on the other hand, found that caregivers were more positive than PWPD themselves when rating the PWPD's communication status. However, the difference was not statistically significant, and it was unclear how much the PWPD and their primary caregivers communicated with one another (Miller et al., 2008). Parveen and Goberman (2017) and Donovan et al. (2008) corroborated

these findings, with the communication partners giving significantly more positive scores for voice handicap and communication effectiveness respectively. Thus, while communication partners, especially those close to or in frequent communication with the individual, serve an important role for PWPD, it is unclear how reliable the judgment of communication partners is with regard to the speech changes in PWPD and how much communication partners' judgement aligns with that of the PWPD themselves.

In other communication disorders such as stuttering, it is recognized that speakers manifest communication breakdowns inherent to their speech difficulties but also evidence associated reactive secondary behaviors, negative thoughts about their speech, and speechrelated anxiety (Vanryckeghem et al., 2004, 2017; Vanryckeghem & Brutten, 2011, 2012, 2018; Wesierska et al., 2018). One instrument used to describe these issues in people who stutter (PWS) is the Behavior Assessment Battery (BAB), an instrument that considers the affective, behavioral, and cognitive reactions associated with communication (BAB, Vanryckeghem & Brutten, 2018). Moreover, this tool has been adapted for use in people with the neurological voice disorder of spasmodic dysphonia (SD), and this version of the instrument is called the Behavior Assessment Battery -Voice (BAB-Voice; Vanryckeghem et al., 2016; Vanryckeghem & Ruddy, 2015; Watts & Vanryckeghem, 2017). The BAB-Voice consists of four subparts: the Behavior Checklist (BCL), the Communication Attitude Test for Adults (BigCAT), the Speech Situation Checklist Emotional Reaction (SSC-ER), and the Speech Situation Checklist Speech Disruption (SSC-SD). The BCL specifically looks at behaviors a speaker might use to deal with his or her voice problem. The BigCAT examines the speaker's attitude, speech-related beliefs, or way of thinking about their voice. The SSC-ER and SSC-SD consider the negative emotional reaction and vocal symptoms

respectively induced by different speech situations. Using the BAB-Voice, speakers with SD consistently showed statistically significant higher scores compared to typical speakers, which indicated elevated anxiety, negative attitude, and significant use of coping behaviors when communicating (Vanryckeghem et al., 2016; Vanryckeghem & Ruddy, 2015; Watts & Vanryckeghem, 2017). Therefore, the BAB-Voice has proven to be a valuable assessment tool for a specific voice disordered population. Given that no other study has extensively measured the attitude, coping mechanisms, and situational emotional reactions and speech disruptions of PWPD, application of the BAB-Voice to PWPD may yield important information to inform our understanding of the psychosocial impact of impaired communication in PWPD. It may potentially inform the development of treatment plans that address more than just the overt perceptual speech and voice disturbances that result from hypokinetic dysarthria. Moreover, in PWS, life partners have been asked to fill out an adapted version of the BAB to investigate if they can perceive the PWS's affective, cognitive, and behavioral reactions to stuttering (Svenning et al., 2021). Svenning et al. (2021) found that the BAB-sub scores of PWS and life partners correlated, though the scores of the PWS were significantly higher for the SSC-ER, and SSC-SD and not significantly different for the BigCAT. Given that the BAB has been adapted to speakers with (neurological) voice disorders, and to allow judgment of communication partners, this is a promising tool to use in a population with PD.

#### Problem Statements, Purposes, Hypotheses & Connections between the Manuscripts

A series of pre-dissertation projects investigated the epidemiological characteristics and treatment outcomes of patients referred to a community voice clinic as well as the auditory-perceptual characteristics of voice production in people with PD at non-advanced stages. The initial study, described in Chapter II, sought to extend epidemiological knowledge from Watts and Knickerbocker (2019) while investigating clinical outcomes of voice treatment within a private practice community voice clinic. The epidemiological patterns of voice disorders in a community voice clinic were similar to those previously reported in laryngologist-led academic medical voice centers. In addition, the measurement of positive treatment outcomes suggests that treatment provided in community voice clinics is effective and comparable to treatment outcomes in other clinical settings. In the subsequent studies, voice disorders, and to a lesser extent, voice treatment, are considered for one specific population: PWPD.

People with Parkinson's disease are among the treatment-seeking populations served by academic medical and community voice clinics. The characteristics of the voice and speech disorders in PWPD during the advanced stages of the disease have been thoroughly described in the literature, including treatment outcomes for those with salient and severe hypophonia. Contrarily, the voice profile and treatment response during the early, nonadvanced stages of PD is less clear. While some acoustic changes likely occur early, the extent to which these voice changes are perceivable is unknown. Therefore, in the second pre-dissertation project (Chapter III) we sought to describe the auditory-perceptual characteristics of voice in PWPD in the non-advanced stages of the disease. This study demonstrated that PWPD were perceived as significantly older, more breathy, and more severely dysphonic than the older healthy controls.

Similar to the second pre-dissertation project on the perceptual voice quality in earlystage PD, the present dissertation study investigates voice disorders in PD, albeit from a novel perspective. The affective and cognitive impact and the coping behaviors related to

communication difficulty in PWPD are not thoroughly understood. Moreover, no other study has extensively measured these affective, behavioral, and cognitive reactions to communication and voice difficulty. Therefore, the purpose of this study was:

- to describe and compare the affective, behavioral, and cognitive reactions to voice disorders in PWPD and healthy controls,
- (2) to describe and compare the affective, behavioral, and cognitive reactions to voice disorders when the tool is administered participant-guided and clinician-guided,
- (3) to describe and compare the affective, behavioral, and cognitive reactions to voice disorders in PWPD as rated by themselves and a daily communication partner,
- (4) to describe how the attitude, emotional reactions, vocal symptoms, and coping behaviors are related to one another in PWPD,
- (5) to determine the internal consistency of the BAB-Voice-items within the different subtests when using it in PWPD.

It was hypothesized (1) that PWPD will present with different affective, behavioral, and cognitive reactions to voice disorders compared to controls, (2) that there will be no difference between administering the instrument via clinician-led or participant-led protocols, (3) that daily communication partners will rate the affective, behavioral, and cognitive reactions to voice disorders as less severe compared to the PWPD themselves,. Similar to what has been described for the BAB-Voice previously, (4) moderate to high correlations between the scoring of the attitude, emotional reactions, vocal symptoms, and coping behaviors on the BAB-Voice subtests were expected, as well as (5) good internal consistency among the items on the different BAB-Voice subtests. This study is described in Chapter IV.

# II. Epidemiological Patterns and Treatment Outcomes in a Private Practice Community Voice Clinic

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#### Abstract

**Objectives**: Voice therapy is administered by speech-language pathologists in multiple practice settings, including private practice community voice clinics. However, the evidence for diagnosis patterns and voice treatment outcomes in community voice clinics is very limited. The purpose of this study was to extend knowledge from a previous investigation by assessing the epidemiological patterns of patient referrals to a private practice community voice clinic across a 4 year period (50 months) and to measure the effectiveness of treatment outcomes for patients who were followed up with voice therapy in that setting.

#### Study Design: Retrospective case series

**Methods**: Consecutive patient records from November 2014 through January 2019 were reviewed. Patients were grouped into 7 categories of distinctive diagnoses. Descriptive data for each group were extracted to determine epidemiological patterns of disorder diagnosis, voice handicap, voice quality severity, age, and gender. For patients who completed at least 3 treatment sessions, pre- and post-treatment measurements of two

assessments, the Voice Handicap Index (VHI) and the Acoustic Voice Quality Index (AVQI), were extracted and compared using a MANOVA.

**Results**: Records from 454 consecutive patient referrals over a 50-month time period were reviewed. The most frequent diagnoses were multifactorial etiologies or those with only a few cases, categorized collectively as an "other" diagnosis category. Diagnoses of nonspecific dysphonia and mid-membranous lesions were also common. CAPE-V scores were not different among disorders, however, group differences were found for VHI and AVQI. Treatment data were available for 292 patients, with 48 of those patients completing at least 3 treatment sessions and with data for pre- and post-therapy VHI and AVQI. A mixed MANOVA showed a significant effect of treatment (Wilks' Lamba=0.42, F[2]=27.58, *P* <0.001,  $\eta_p^2$ =0.58), where both AVQI and VHI improved significantly across the pre-to-post treatment measurements.

**Conclusion**: Patient characteristics and diagnosis patterns across a 50 month period were similar when compared to a previous study that investigated epidemiological patterns in this clinic across 28 months. Voice therapy administered in this community voice clinic to patients with varied diagnoses was found to be effective based on changes in VHI and AVQI measurements.

**Keywords:** Voice therapy – private practice voice clinic – effectiveness – voice quality – perceived voice handicap

#### Introduction

Voice therapy is often implemented as a primary treatment approach for voice disorders and can, in some cases, change subsequent recommendations for medical management (Desjardins et al., 2017; LeBorgne & Donahue, 2019). While the majority of evidence reporting voice treatment outcomes and epidemiological data for patient populations with voice impairments has been from laryngologist-led clinics typically associated with academic medical centers (Cohen et al., 2012, 2015), a substantial number of speech-language pathologists (SLPs) who are voice clinicians see patients in community voice clinics that are not led by a physician (Watts & Knickerbocker, 2019). For example, as of the year 2020 more than 8,600 SLPs identify "voice" as one of their clinical specialty areas, with over 550 of these professionals working in nonacademic community settings such as private offices or speech clinics (American Speech-Language-Hearing Association, 2020).

Regardless of practice setting, a commonly used tool for assessing the outcomes of voice therapy is the Voice Handicap Index (VHI) (Jacobson et al., 1997). As a patient-reported outcome measure, the VHI represents a primary tool used to measure treatment outcomes in patients receiving voice therapy (Francis et al., 2017; Watts et al., 2019). The VHI consists of 30 questions divided over three domains (emotional, physical, and functional subscales) (Jacobson et al., 1997). All items are scored on a Likert scale ranging from 0—never to 4—always, resulting in a score between 0 and 120. Higher scores on the VHI reflect a greater perceived vocal handicap (Jacobson et al., 1997). The VHI's clinical utility is supported by robust psychometric properties compared to other patient-reported quality of life instruments (Franic et al., 2005). Moreover, the administration of the VHI is standardized which has allowed for comparisons of scores across a large body of clinical research

(Kapsner-Smith et al., 2015; Roy et al., 2003; Watts et al., 2019; Watts, Diviney, et al., 2015; Watts, Hamilton, et al., 2015).

Acoustic assessments of vocal function are also a standard component of a comprehensive voice evaluation and have been shown to be sensitive to treatment change in populations with voice impairments (Alharbi et al., 2019; Maryn, De Bodt, et al., 2010). One example is the Acoustic Voice Quality Index (AVQI), a multiparametric acoustic measure that is both strongly related to perceptions of voice quality (eg, perceived severity of dysphonia) and sensitive to treatment outcomes (Maryn, De Bodt, et al., 2010). The AVQI can be used with the computer program Praat (Boersma & Weenink, 2020) and combines multiple cepstral, spectral, and time-based measurements applied to continuous speech and sustained vowel recordings. AVQI analysis produces a single metric that falls within a continuum of 1 to 10, where higher numbers are associated with greater dysphonic severity (Maryn, Corthals, et al., 2010; Watts & Awan, 2019). The use of the AVQI as a diagnostic tool and as a measure of clinical outcomes is supported by a large and growing body of evidence across multilinguistic patient populations (Kankare et al., 2019; Maryn et al., 2014; Pommée et al., 2018).

It stands to reason that a large population of patients with voice disorders is evaluated and treated in community voice clinic settings. Unfortunately, we know very little about the demographics and characteristics of the populations referred to private practice clinics, and even less about the outcomes of the voice therapy administered to patients treated in these settings. Our knowledge of epidemiological patterns in different clinical practice sites is important for at least three reasons: (1) for benchmarking referral patterns and caseload characteristics that will inform what practices the clinician will need to utilize in their

professional role (ie, what evaluation tools and skills are needed, what treatment approaches should the clinician be competent in, etc?); (2) for monitoring trends in incidence and prevalence of voice disorders over time in specific practice settings and communities (ie, disease surveillance); and (3) to provide evidence of epidemiological patterns that can inform future studies seeking to identify risk factors for voice disorders in different populations (ie, is gender represented disproportionately, is there a large percentage of certain professions represented across clinical settings?). In a recent study, we addressed this problem by reporting epidemiological data from 216 patients evaluated in a private practice community voice clinic across a 28-month period (Watts & Knickerbocker, 2019). That study found that patient demographics and diagnosis distributions in a private practice clinic led by an SLP were similar to those in specialty voice clinics led by laryngologists. It was concluded that the competencies needed by SLPs in a private clinic would be the same as SLPs working in specialty voice centers. This is critical knowledge, as it should inform the educational and experiential needs of clinicians considering voice/vocology as a specialty area, regardless of practice setting.

The purpose of this study was to extend our previous research using the following research questions: (1) what are the epidemiological patterns in a private practice community voice clinic including a large sample of patients evaluated across 50 consecutive months; and (2) what are voice treatment outcomes as measured by the VHI and AVQI tools for patients treated in the same private practice setting. For the second research question, we hypothesize that treatment outcomes after at least three voice therapy sessions will show measurable and significant decreases in VHI and AVQI scores, and thus show that voice therapy intervention can result in positive outcomes in a private practice setting.

#### Methodology

This study was a continuation of our previous retrospective investigation of diagnosis and referral patterns to a private practice community voice clinic (Watts & Knickerbocker, 2019). The study was approved by the Texas Christian University Institutional Review Board. Epidemiological data were collected from all patients receiving a voice evaluation across 50 consecutive months (28 months from earlier study, 22 additional months for new data set) and their patient charts in a single clinical practice. The data represented patients referred to a single community voice clinic, which was led by a licensed and certified SLP (second author, KK) with a background in vocal performance and 7 years of clinical experience centered on vocology. All patients referred to the clinic were included in the sample, regardless of age and diagnosis.

All patient referrals to the voice clinic were from either (1) community otolaryngology practices, (2) community SLPs, or (3) self-referrals. Data extracted for this investigation represent information obtained during a specialty voice evaluation by the SLP, along with records from the patients' otolaryngologist. Previous or subsequent otolaryngology examination was completed for each patient prior to the development of any voice treatment plan. Medical diagnoses were established via the otolaryngology and specialty voice clinical evaluations. Laryngeal imaging via videostroboscopy was completed by either a referring otolaryngologist or the SLP. The International Classification of Diseases, 10th Revision was utilized for guidance on diagnosis codes, consistent with previous studies (Roy et al., 2016; Watts & Knickerbocker, 2019).

Within this overall sample, we also identified patient charts that represented those who underwent an initial evaluation and received at least three sessions of voice therapy in the clinic, and for whom pretreatment and posttreatment data were available to evaluate treatment outcomes in this private practice community voice clinic. When a voice treatment plan was developed for a patient who would be served by the clinic, voice therapy was personalized for each patient and was based on their diagnosis and needs. Treatment plans consisted of one or more of the following domains: voice production physiology education, vocal wellness education (ie, vocal hygiene), and a combination of Resonant Voice Therapy, semioccluded vocal tract exercises, Stretch-and-Flow and/or Vocal Function Exercises. All patients were treated individually and in-person during weekly therapy sessions. If needed, adjacent medical management was sought. Patients were discharged after typically 3-5 therapy sessions if they met one of the following criteria: (1) the patient no longer demonstrated vocal impairment, and/or (2) voice therapy goals were met; and/or (3) the patient was able to apply new vocal behaviors confidently and independently to their satisfaction.

The following data were extracted from the charts by the treating SLP (second author, KK) for all patients: (1) voice disorder diagnosis, (2) age, (3) gender, (4) auditory perceptual ratings of voice quality, (5) AVQI scores, and (6) VHI scores. The auditory perceptual ratings of voice quality were performed by the SLP using the Consensus Auditory Perceptual Evaluation of Voice-scale (CAPE-V) (Kempster et al., 2009). The disorder diagnoses were stratified into different categories, using a similar approach as our previously published study (Watts & Knickerbocker, 2019). This resulted in eight distinct groups of diagnoses: (1) atrophy or bowing, (2) midmembranous lesions [MMLs], (3) muscle tension dysphonia [MTD], (4) nonspecific dysphonia [NSD], (5) patients who are transgender, (6) vocal cord dysfunction [VCD], (7) vocal fold immobility [paresis or paralysis – VFI], and (8) an

"Other" category consisting of etiologies that represented less than five patients, patients with multifactorial diagnoses, patients diagnosed with "acute laryngitis," or patients with no divergent voice quality. For the subgroup of patients for whom pretreatment and posttreatment data were available, we chose to use pretreatment and posttreatment measurements of VHI and AVQI as the outcome measures in this study, as these metrics can be validly compared within and across patients. Therefore, the posttreatment measurements for VHI and AVQI were extracted from the patient files as well for these patients.

We applied multivariate analyses of variance (MANOVA) to the VHI, CAPE-V, and AVQI scores for the entire data set with diagnosis group (etiology) as the main factor. MANOVA analysis was selected because it allowed for the inclusion of variance associated with all three dependent variables in one statistical test, and in doing so served to protect against Type 1 error for any follow-up tests to the MANOVA (Field, 2013). For all statistical analyses, a significance level of  $\alpha = 0.05$  was set. To examine the epidemiological patterns of all patients referred to the clinic, we merged data sets from the 22-month cohort (new data) with the 28-month cohort (previously published data). A single data set representing patients evaluated in the voice clinic across 50 consecutive months was derived. Descriptive analyses were then applied to this pooled data set, containing all patients, to describe the following epidemiological patterns: (1) diagnosis category frequency, (2) age distribution, (3) gender distribution. For statistically significant main effects, post hoc testing utilized the Fisher least square difference test in a pairwise manner. To determine the outcome of voice treatment, we only considered the patients for whom pretreatment and posttreatment data were available. A MANOVA was also applied to this data, with diagnosis group (etiology) and measurement period (pretreatment vs posttreatment) as factors. Pairwise testing using the Fisher least

square difference test for statistically significant main effects was used for post hoc testing. For all inferential analyses, outliers outside 1.5x the interquartile range were removed, as defined by the statistical program (IBM SPSS, v. 25).

#### Results

#### **Research Question 1: Characteristics of the Whole Patient Population**

Of the reported 454 patients that were referred to the community voice clinic for initial evaluation, 225 cases were presented in our previous data set (Watts & Knickerbocker, 2019), and 229 cases were reported for the first time. As no significant differences between the groups were found, the groups were pooled together. An overview of the diagnoses and patient characteristics of the 454 patients can be found in Figure 1 and Table 1. The most prevalent disorder diagnosis across the sample was "Other" (25.3%) followed by NSD (24.7%) and MMLs (15.4%). More females (67.4%) were seen in the clinic compared to males (32.6%). Moreover, Table 1 shows that the sample contained more patients in older age groups compared to younger age groups. The most common diagnoses in females were the "Other" category and NSD (both 16.7%), followed by MMLs (12.3%). In males, the "Other" diagnosis and NSD were the most prevalent, at 8.6% and 7.9%, respectively. Patients who are transgender were the third most prevalent category (5.2%) for the male gender in this clinical practice. Table 2 shows the average age, CAPE-V overall severity score, as well as the average VHI and AVQI score for each diagnosis group in the full sample. The diagnoses atrophy and bowing were most prevalent in older patients, whereas patients who are transgender were the youngest. CAPE-V scores were similar across all groups. The AVQI value for VCD appeared to be higher (indicating worse voice quality) than the other groups.

The CAPE-V, VHI, and AVQI scores of the patient sample at the initial evaluation were compared using a MANOVA with diagnosis group as a between-subject factor and the three measurements as separate variables in the statistical model. Twenty data points in the VHI data set were identified as outliers and removed along with six data points from the AVQI data. The analysis showed a significant effect for diagnosis group (Wilks' Lambda = 0.842, F[21] = 2.78, P < 0.001). Significant group differences were found for VHI (F[7] =4.20, P < 0.001), and AVQI (F[7] = 3.21, P = 0.003), but not for CAPE-V score (F[7] = 1.05, P = 0.39). Post hoc tests revealed that for VHI, patients with MMLs scored significantly higher than patients with NSD (P < 0.001), patients who are transgender (P = 0.011), and the "other" diagnosis group (P = 0.002). Patients with VFI also scored significantly higher than patients with NSD (P < 0.001), patients who are transgender (P = 0.036), and the "Other" diagnosis group (P = 0.020). Patients with atrophy or bowing showed significantly higher VHI scores than the NSD group (P = 0.026). For AVQI, patients with VFI scored significantly higher than MMLs (P = 0.014), NSD (P = 0.002), patients who are transgender (P = 0.001), and patients with MTD (P = 0.005). Patients with atrophy or bowing also had higher AVQI scores than patients who are transgender (P = 0.007) and patients with MTD (P= 0.022). The "Other" category also had significantly higher scores than patients who are transgender (P = 0.018).

## Table 1

Cross-tabulation of Disorder Diagnosis, Age Categories, and Gender of the Whole Sample (Transgender Patients Are

Coded Based on Biological Gender)

Diagnosis	Gender	<20	20-29	30-39	40-49	50-59	60-69	≥70	Total
MML	Male	1 (0.2%)	0 (0%)	2 (0.4%)	5 (1.1%)	2 (0.4%)	2 (0.4%)	2 (0.4%)	14 (3.1%)
	Female	4 (0.9%)	9 (2.0%)	14 (3.1%)	15 (3.3%)	10 (2.2%)	3 (0.7%)	1 (0.2%)	56 (12.3%)
	Total	5 (0.11%)	9 (2.0%)	16 (3.5%)	20 (4.4%)	12 (2.6%)	5 (0.11%)	3 (0.7%)	70 (15.4%)
VFI	Male	1 (0.2%)	1 (0.2%)	0 (0%)	4 (0.9%)	0 (0%)	4 (0.9%)	5 (1.1%)	15 (3.3%)
	Female	2 (0.4%)	1 (0.2%)	2 (0.4%)	9 (2.0%)	11 (2.4%)	11 (2.4%)	15 (3.3%)	51 (11.2%)
	Total	3 (0.7%)	2 (0.4%)	2 (0.4%)	13 (2.9%)	11 (2.4%)	15 (3.3%)	20 (4.4%)	66 (14.5%)
Atr	Male	0 (0%)	0 (0%)	0 (0%)	1 (0.2%)	1 (0.2%)	5 (1.1%)	8 (1.8%)	15 (3.3%)
	Female	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	6 (1.3%)	12 (2.6%)	18 (4.0%)
	Total	0 (0%)	0 (0%)	0 (0%)	1 (0.2%)	1 (0.02%)	11 (2.4%)	20 (4.4%)	33 (7.3%)
NSD	Male	1 (0.2%)	4 (0.9%)	2 (0.4%)	7 (1.5%)	3 (0.7%)	10 (2.2%)	9 (1.9%)	36 (7.9%)
	Female	3 (0.7%)	9 (2.0%)	6 (1.3%)	8 (1.8%)	22 (4.8%)	14 (3.1%)	14 (3.1%)	76 (16.7%)
	Total	4 (0.9%)	13 (2.9%)	8 (1.8%)	15 (3.3%)	25 (5.5%)	28 (6.1%)	23 (5.1%)	112 (24.7%)
VCD	Male	0 (0%)	0 (0%)	1 (0.2%)	1 (0.2%)	0 (0%)	0 (0%)	1 (0.2%)	3 (0.7%)
	Female	5 (1.1%)	0 (0%)	2 (0.4%)	3 (0.7%)	1 (0.2%)	1 (0.2%)	0 (0%)	12 (2.6%)
	Total	5 (1.1%)	0 (0%)	3 (0.7%)	4 (0.9%)	1 (0.2%)	1 (0.2%)	1 (0.2%)	15 (3.3%)
Trans	Male	7 (1.5%)	8 (1.8%)	7 (1.5%)	1 (0.2%)	1 (0.2%)	0 (0%)	0 (0%)	24 (5.3%)
	Female	0 (0%)	0 (0%)	1 (0.2%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	1 (0.2%)
	Total	7 (1.5%)	8 (1.8%)	8 (1.8%)	1 (0.2%)	1 (0.2%)	0 (0.0%)	0 (0%)	25 (5.5%)
MTD	Male	0 (0%)	0 (0%)	1 (0.2%)	0 (0%)	0 (0%)	1 (0.2%)	0 (0%)	2 (0.4%)
	Female	2 (0.4%)	2 (0.4%)	1 (0.2%)	5 (1.1%)	1 (0.2%)	2 (0.4%)	3 (0.7%)	16 (3.5%)
	Total	2 (0.4%)	2 (0.4%)	2 (0.4%)	5 (1.1%)	1 (0.2%)	3 (0.7%)	3 (0.7%)	18 (4.0%)
Other	Male	2 (0.4%)	3 (0.7%)	4 (0.9%)	5 (1.1%)	11 (2.4%)	7 (1.5%)	7 (1.5%)	39 (8.6%)

	Female	2 (0.4%)	2 (0.4%)	4 (0.9%)	9 (2.0%)	12 (2.6%)	27 (5.9%)	20 (4.4%)	76 (16.7%)
	Total	4 (0.9%)	5 (1.1%)	8 (1.8%)	14 (3.1%)	23 (5.1%)	34 (7.5%)	27 (5.9%)	115 (25.3%)
Total	Male	12 (2.6%)	16 (3.5%)	17 (3.7%)	24 (5.3%)	18 (4.0%)	29 (6.4%)	32 (7.0%)	148 (32.6%)
	Female	18 (4.0%)	23 (5.1%)	30 (6.6%)	49 (10.8%)	57 (12.6%)	64 (14.1%)	65 (14.3%)	306 (67.4%)
	Total	30 (6.6%)	39 (8.6%)	47 (10.4%)	73 (16.08%)	75 (16.5%)	93 (20.5%)	97 (21.4%)	454 (100%)

Abbreviations: MML, mid-membranous lesion; VFI, vocal fold immobility; Atr/Bow, atrophy or bowing; NSD, non-specific

dysphonia; VCD, vocal cord dysfunction; Trans, patient who is transgender; MTD = muscle tension dysphonia.

### Figure 1

Pie Chart Describing the Prevalence of Each Diagnosis Across the Whole Sample.



*Note*. MML, mid-membranous lesion; VFI, vocal fold immobility; Atr/Bow, atrophy or bowing; NSD, non-specific dysphonia; VCD, vocal cord dysfunction; Trans, patient who is transgender; MTD, muscle tension dysphonia.
#### Table 2

	Age	VHI	CAPE-V	AVQI
MML	42.76 (14.71)	39.20 (27.37)	50.44 (25.03)	3.90 (1.68)
VFI	58.74 (16.25)	36.64 (23.11)	50.94 (26.65)	4.72 (2.00)
Atr/Bow	74.57 (8.12)	33.07 (15.39)	49.00 (23.18)	4.50 (1.57)
NSD	54.30 (18.82)	22.72 (16.65)	41.22 (25.98)	3.76 (1.60)
VCD	31.50 (24.75)	39.00 (16.97)	46.50 (45.96)	5.20 (0.22)
Trans	28.29 (10.12)	24.18 (21.53)	52.41 (35.85)	3.00 (1.10)
MTD	45.55 (14.46)	34.30 (22.43)	41.00 (27.04)	2.99 (0.97)
Other	57.93 (16.36)	27.94 (20.92)	45.20 (29.88)	4.13 (2.09)
Total	52.46 (19.40)	30.45 (21.98)	46.58 (27.42)	4.04 (1.81)

Means and Standard Deviations of Quantitative Variables for Each Diagnosis Group

(maximal perceived handicap). CAPE-V scores range between 0 (no deviation from normal) and 100 (maximal deviation from normal). AVQI score range from 0 (good voice quality) to 10 (bad voice quality).

Note. Age is reported in years. VHI scores range between 0 (no perceived handicap) and 120

Abbreviations: MML, mid-membranous lesion; VFI, vocal fold immobility; Atr/Bow,

atrophy or bowing; NSD, non-specific dysphonia; VCD, vocal cord dysfunction; Trans,

patient who is transgender; MTD, muscle tension dysphonia.

#### **Research Question 2: Voice Treatment Outcomes**

Across the entire data set, 292 patients were referred for and did attend at least one voice therapy session. Criteria for treatment outcome analysis (completed a minimum of three treatment sessions, and pre- and posttreatment data were available for VHI and AVQI) were met for 54 of these cases (12%). Patients with VCD and those who were transgender were not included in the analysis as dysphonia was not their main complaint. Those with MTD (ie, nonphonotraumatic dysphonia) were collapsed into the "Other" group due to

insufficient numbers. Seven cases were identified as outliers and removed from the analysis. Therefore, 47 patients were included in the analysis. An overview of the average scores for AVQI and VHI pre- and posttreatment can be found in Table 3. The pretreatment scores for both VHI and AVQI appeared to be numerically higher than the posttreatment scores. The MMLs group showed the highest VHI and AVQI score, whereas NSD showed the lowest scores.

The differences between the pretreatment and posttreatment values for the disorder diagnosis groups were calculated using a 2 x 5 Mixed MANOVA design with measurement time (pretreatment vs posttreatment) and diagnosis group as the factors where VHI and AVQI scores were variables in the statistical model. There was only a significant effect of time, ie, pretreatment measurements vs posttreatment measurements (Wilks' Lambda = 0.42, F[2] = 27.58, P < 0.001,  $\eta_p^2 = 0.58$ ). No significant effect of group (Wilks' Lambda = 0.68, F[10] = 1.73, P = 0.09,  $\eta_p^2 = 0.18$ ) was found. No interaction effect between diagnosis group and time was found (Wilks' Lambda = 0.80, F[10] = 0.93, P = 0.51,  $\eta_p^2 = 0.10$ ). The withinsubject analyses showed that pretreatment VHI scores were significantly higher than the posttreatment VHI scores (F[1] = 35.02, P < 0.001,  $\eta_p^2 = 0.43$ ) when collapsing across all diagnosis groups. That is, across all 47 patients the average VHI and AVQI scores decreased, indicating an improvement in perceived handicap and acoustic indices of voice quality (Figures 2 and 3).





*Note.* The scores pre-therapy are represented by the left, blue boxplots, while the posttherapy scores are the right, red boxplots. The upper line of the colored box represents the third quartile (percentile 75), the middle line represents the median, while the lower line represents the first quartile (percentile 25). The ends of the whiskers designate the minimum and maximum values within 1.5x the interquartile range.

*Abbreviations:* VHI, Voice Handicap Index; MML, mid-membranous lesion; VFI, vocal fold immobility; Atr/Bow, atrophy or bowing; NSD, non-specific dysphonia; VCD, vocal cord dysfunction; Trans, patient who is transgender; MTD, muscle tension dysphonia.



Boxplots of the AVQI-scores Pretreatment and Poststreatment

*Note.* The scores pre-therapy are represented by the left, blue boxplots, while the posttherapy scores are the right, red boxplots. The upper line of the colored box represents the third quartile (percentile 75), the middle line represents the median, while the lower line represents the first quartile (percentile 25). The ends of the whiskers designate the minimum and maximum values within 1.5x the interquartile range.

*Abbreviations:* AVQI, Acoustic Voice Quality Index; MML, mid-membranous lesion; VFI, vocal fold immobility; Atr/Bow, atrophy or bowing; NSD, non-specific dysphonia; VCD, vocal cord dysfunction; Trans, patient who is transgender; MTD, muscle tension dysphonia.

#### Table 3

Means and Standard Deviations for Pre- and Posttreatment Scores for Each Group Included in the Analysis.

Diagnosis	n	Pre VHI	Post VHI	Pre AVQI	Post AVQI
ML	9	49.78 (28.91)	21.89 (19.10)	4.60 (1.74)	3.43 (1.29)
VFI	8	34.12 (18.61)	6.50 (5.73)	4.25 (1.38)	3.44 (1.61)
Atr/Bow	7	32.71 (8.98)	12.57 (8.83)	5.10 (1.11)	3.18 (1.28)
NSD	10	20.60 (18.61)	4.70 (3.92)	4.06 (1.48)	2.43 (1.14)
MTD	3	45.00 (33.78)	34.00 (34.40)	4.17 (0.58)	2.88 (0.91)
Other	10	30.20 (14.20)	21.00 (16.17)	4.28 (2.27)	2.96 (0.81)
Total	47	33.89 (21.55)	14.81 (16.17)	4.40 (1.60)	3.05 (1.20)

*Note*. VHI scores range between 0 (no perceived handicap) and 120 (maximal perceived handicap). AVQI score range from 0 (good voice quality) to 10 (bad voice quality). *Abbreviations:* MML, mid-membranous lesion; VFI, vocal fold immobility; Atr/Bow, atrophy or bowing; NSD, non-specific dysphonia.

## Discussion

The purpose of this study was to further investigate diagnosis and referral patterns in a private practice community voice clinic led by an SLP, as well as describing treatment outcomes after at least three voice therapy sessions. We compiled data from a group of 454 patients seen across a consecutive 50-month period. We analyzed the epidemiological characteristics of this group and also treatment outcomes for a subgroup of patients who received at least three sessions of voice therapy. Our main findings were as follows: (1) Across all 454 patients evaluated in the voice clinic, two thirds were females; (2) 50% of all disorder diagnoses were represented by the "Other" category (within this category the most common diagnosis was acute laryngitis) and the NSD category; (3) The disorder diagnoses of MMLs, VFI, and atrophy/bowing had the largest negative effects on VHI and AVQI scores; (4) Of 47 patients included in the pre- to posttreatment analyses, there was a significant treatment effect on measurements of VHI and AVQI, suggesting positive treatment outcomes on measures of voice handicap and acoustic measures of voice quality. However, voice therapy did not result in the same degree of improvement for these VHI or AVQI measures across the different diagnosis groups.

The current study was a continuation of a previously published investigation that reported the diagnosis and referral patterns in a private practice community voice clinic. While some minor differences were observed (most noticeably an increase in the percentage of NSD and "Other" cases), the diagnosis patterns were largely the same in the new group of 229 patients compared to the same measurements from our previously reported data set of 225 patients. The epidemiological patterns of gender, age, and VHI scores, and CAPE-V ratings were also similar in the two cohorts and no significant differences were present between the data sets. The sample reported in this study, which combines both groups of data, therefore did not appear to have changed over time. Moreover, the overall demographic characteristics of our current sample were largely similar to what has been reported in the literature. The current sample found that 67% of the patients evaluated in the voice clinic were female. Previous studies have reported very similar percentages, ranging between 60% and 70% (Coyle et al., 2001; De Bodt et al., 2016; Misono et al., 2016; Mozzanica et al., 2016; Remacle et al., 2017; Van Houtte et al., 2010). These gender differences have been explained previously as due to dissimilarities in laryngeal anatomy between males and

females, where anatomical and resulting physiological differences put females at a greater risk for specific voice disorders (Cohen et al., 2012; Coyle et al., 2001; Misono et al., 2016; Van Houtte et al., 2010). Moreover, our sample showed similar age distributions as previous studies: over half of the patients in our sample were over 50 years old (Coyle et al., 2001; Mozzanica et al., 2016; Van Houtte et al., 2010). However, some studies have reported noticeably younger populations (De Bodt et al., 2016; Remacle et al., 2017), possibly because of differences in the clinic settings from which epidemiological data were collected.

The current study found that the "Other" category (25.33%) and NSD (24.66%) were the most prevalent disorders, followed by MMLs (15.4%) and VFI (14.54%), which is in accordance with our previously published report from approximately one-half of the current data set (Watts & Knickerbocker, 2019). As the "Other" category in the current study included multidimensional diagnoses (patients with multiple diagnoses, diagnoses with less than five cases, etc) it is difficult to directly compare this category with previous literature. However, the three most prevalent diagnoses besides the "Other" category were very similar to what has been reported in published studies. Vocal fold nodules are often found to be one of the most prevalent diagnoses within a sample in a voice clinic. Reported percentages of patients presenting with vocal fold nodules have ranged between 10% and 23% (Coyle et al., 2001; De Bodt et al., 2016; Mozzanica et al., 2016; Remacle et al., 2017; Van Houtte et al., 2010), which is comparable to the 15% we found in the current sample, although we also included other MMLs, such as vocal fold cysts and polyps, in this group. Second, VFI (vocal fold paralysis or paresis) has been reported to occur in 8% to 24% of patient populations (Coyle et al., 2001; De Bodt et al., 2016; Misono et al., 2016; Mozzanica et al., 2016;

Remacle et al., 2017; Van Houtte et al., 2010), which is also comparable to our findings (14.45%).

NSD, defined in this study as patients without observable laryngeal impairment or obvious functional components such as MTD, was the diagnosis in 24.66% of the cases. While the terminology within the literature differs, percentages between 8% and 12% have been reported previously for similar diagnosis categories. Some studies found a markedly higher prevalence of MTD than the current study (De Bodt et al., 2016; Mozzanica et al., 2016; Van Houtte et al., 2010). The discrepancy between the current study and previously reported literature for NSD and MTD may stem from differences in definitions and methodology. For patients diagnosed with atrophy, VCD, MTD, and patients who are transgender the prevalence was low, which is consistent with the literature (Misono et al., 2016; Remacle et al., 2017; Van Houtte et al., 2010). It is important to note that all of the previously published literature reported data from academic medical centers or clinics led by ENTs. Our findings continue to support the supposition that clinical populations in an SLPled private practice voice clinic are similar to patient populations in physician-led voice clinics. The results of our study did differ from data reported from insurance claims. Those studies have found acute laryngitis to be the most prevalent disorder diagnosis (42%-54%), followed by NSD (22%-31%) (Benninger et al., 2017; Cohen et al., 2014).

The current study found a significant positive effect of treatment for patients attending a private practice voice clinic. No significant differences were found among the diverse diagnoses. Therefore, the current study does not support the notion that the effect of therapy on voice disorders was dependent on the disorder type. However, our study sample included in the pretreatment and posttreatment comparison was small: forty-seven

participants divided over six groups of diagnoses. Studies with larger samples could provide a more representative indication of therapy outcomes across diagnosis groups. Another possible explanation is within-group variability. The "Other" group contained patients with multiple diagnoses as well as diagnoses with low prevalence. It is possible these different pathologies reacted differently to therapy and thus minimalized the effect of therapy within the group. Similarly, the MMLs group contained patients with nodules, cysts, and polyps. Ogawa and Inohara (2018) described the differential effects of treatment for patients with vocal fold polyps, nodules, and cysts. More specifically, cysts would not respond well to voice therapy (Ogawa & Inohara, 2018). Therefore, combining cysts with more manageable MML may have impacted the group's therapy outcome in this study.

The outcome of therapy was measured with the VHI and AVQI. We chose VHI as a treatment outcome measure in this study because this tool measures the patient's perception of their own voice problem and its ubiquitous use in diagnostic and treatment studies across the vocology and laryngology literature. Pre- to posttreatment changes of 18 points or greater in VHI scores are considered a meaningful treatment response for perceived vocal handicap (Jacobson et al., 1997). Rosen et al (2000) and Bouwers et al (2009) found lower VHI scores after treatment in patient populations, and there were similar degrees of improvement in VHI scores across the two studies. However, both studies took place in an academic voice center and had surgery as one of the treatment options (Bouwers & Dikkers, 2009; Rosen et al., 2000). In the present study, posttreatment VHI scores for patients with MMLs, VFI, and atrophy or bowing all improved beyond the 18-point threshold. This finding supported our assumption that treatment administered by a clinician experienced in treating voice disorders in a private practice community voice clinic can be effective for reducing vocal handicap.

The three diagnosis categories that did not improve by 18 VHI points or more included NSD, MTD, and the "Other" category. Although VHI treatment changes in patients within both of those diagnosis groups were statistically significant, the failure of those changes to reach the 18 point threshold may be due to (1) the nonspecificity of the underlying impairment for the NSD group, which presented a barrier for developing targeted treatment plans that addressed a specific physiological imbalance, (2) the small number of patients with MTD in the sample, and (3) the heterogeneity of patients within the "Other" group, which included those with acute laryngitis, chronic cough, and VCD among several other diagnoses.

The results of our investigation indicated that voice therapy significantly decreased AVQI measurements, indicating improvement, when comparing pre- and posttreatment values. AVQI has previously shown to be an effective tool for measuring acoustic indices of voice quality that change secondary to voice therapy (Maryn, De Bodt, et al., 2010). The positive change in AVQI measures from our study aligns with findings from Chhetri and Gautam (2015), who also found improvement in acoustic voice measures secondary to treatment. Meerschman et al (Meerschman et al., 2019) also found improved AVQI scores, although the reported degree of improvement was not statistically significant in their traditional voice therapy group. In other trials with more specific therapy approaches, AVQI is often used as an outcome measure and has been shown to be sensitive to improved vocal function after voice therapy (Barsties v. Latoszek, 2020; Rubin et al., 2019; Watts et al., 2019).

Interestingly, when using a cut-off score of 2.95 for the AVQI to distinguish normal from disordered voice quality (Maryn, De Bodt, et al., 2010), not all groups had an objective "normal" voice at voice treatment discharge. Patients in the diagnosis groups with VFI,

MMLs, and atrophy or bowing still manifested acoustic measures above the AVQI threshold of normal. It is important to note that this threshold value has been validated in a Dutchspeaking population only, whereas the population evaluated in the clinic of this study were English speaking and the vast majority spoke English as a first language. The deviation of the patients' voices from normal at discharge could be explained by multiple factors. First, VFI, MMLs, and atrophy or bowing are all three organic pathologies of the vocal folds. For these etiologies, voice treatment alone may not be sufficient to restore normal voice quality in a number of patients, who may need further medical treatment (Carding et al., 2017; Walton et al., 2018). Ogawa and Inohara (2018) have found mixed results for voice therapy in people with benign vocal fold lesions. Moreover, in patients with VFI, the effect of voice therapy as a primary treatment remains unclear (Walton et al., 2018). For vocal fold bowing and/or atrophy, voice therapy by itself might only be effective in mild cases (Kost & Sataloff, 2018).

Second, dysphonic voice quality at discharge could be explained by the different criteria set for discharge when developing individualized voice treatment plans for specific patients. Normal voice quality is not the primary goal for the patient or voice therapist. Gillespie and Gartner-Schmidt (2018) found that the five most important criteria for the discharge of a patient treated for a voice disorder were (1) independently using the new voice, (2) being able to function in daily life with their new voice, (3) being able to differentiate the good from the bad voice as well as (4) taking responsibility for their voice, and (5) a better sounding voice than pretreatment. These authors suggested that for discharge, the patient's ability to generalize their skills to daily life is considered to be more important than acoustic outcomes (Gillespie & Gartner-Schmidt, 2018). Similar goals were reflected in the discharge criteria for many of the 47 patients included in the analysis, and could therefore further explain why AVQI scores for some patients were above the normal threshold.

Most studies that investigate therapy outcomes or effectiveness either focus on a specific population and/or a specific therapy technique within a fixed time frame and a controlled setting (Desjardins et al., 2017). While this methodology is ideal to determine the effectiveness of therapy in these populations or with those techniques, it does not reflect typical speech-language pathology practice patterns. Multiple authors have recognized that SLPs use multiple direct voice therapy techniques in practice to accommodate a patient's needs (Burg et al., 2015; Chan et al., 2013; Gartner-Schmidt et al., 2013). The value of the current study is, therefore, that it looked at voice therapy outcomes in an ecologically valid manner. The patients were treated using an individualized treatment plan that often comprised multiple voice therapy techniques, which is reflective of real-world clinical practice. The current study provides support for the notion that voice therapy is effective, even when multiple therapy techniques are employed, and when those specific combinations of techniques are different for each patient. Our findings thus suggest that voice therapy can be effective in a private practice community voice clinic led by an SLP.

It is important to note that a substantial number of the patients evaluated in the discussed private practice community voice clinic did not undergo voice therapy. There are several barriers to the implementation of voice therapy in treatment-seeking populations: the patient may not want to participate in treatment because he/she did not understand the purpose of therapy; the patient might believe that behaviors targeted in voice therapy will not translate to daily life; the patient may perceive the exercises targeted in therapy are strange or hard; or because the patient wants to wait to see if the voice impairment will spontaneously

recover (Misono et al., 2017). Van Leer and Connor (2010) have concluded that voice therapy requires substantial resources of the patient, including motivation, self-regulation, and the ability to form a productive relationship with the therapist. Moreover, insurance coverage and travel requirements have also been reported as patient barriers to participation in voice therapy (Portone et al., 2008).

The current retrospective study design presented a number of limitations that must be considered. First, treatment plans were individualized based on the patient's needs and goals, and as such the specific therapeutic approaches used were not identical from patient to patient. While this reflects real-world clinical practice, it did not allow us to compare one treatment approach or technique to another. We were unable to determine if voice treatment outcome was influenced by medical or surgical treatment, as not all patient records available in the private practice clinic had surgical notes if the patient previously underwent surgical intervention. Moreover, the data for this study were gathered by the SLP who treated the patients. This could bias the findings of the study, as the therapist was not blinded for the diagnosis of the participant. Another limitation is that voice therapy outcome data were available for only 292 patients, of which only 47 could be included in the treatment outcomes analysis. A reason for this is that posttreatment acoustic and VHI data were not always collected at the moment of therapy discharge for each patient seen in this clinic. This resulted in a limited sample size for that part of the analysis. To address the limitations noted above, future research can utilize a prospective methodology to better control for factors that influence data collection and treatment outcomes.

# Conclusion

The current study aimed to investigate the epidemiological patterns as well as the effectiveness of voice therapy for patients referred to a private practice community voice clinic. The patient characteristics and diagnosis patterns found in the current study were comparable to previously reported data from a smaller sample of the same population. Across the whole sample, diagnoses in the "Other" category, MMLs, and NSD were the most common. The different diagnoses presented with diverse VHI and AVQI ratings, although the CAPE-V ratings were similar across diagnoses. For 47 patients who received at least three sessions of voice therapy, measures of VHI and AVQI improved significantly at posttreatment, although the degree of improvement was not the same for all disorder diagnosis categories. Collectively, the results of this study indicated that voice therapy in a community private practice voice clinic is effective for improving acoustic voice measures and self-perceived voice handicap.

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# III. Perceptual Characterization of Voice Quality in Non-Advanced Stages of Parkinson's Disease

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#### Abstract

**Introduction**: Parkinson's disease is a neurodegenerative disorder that impacts motor and non-motor systems, and consequently influences voice. In later stages of the disease, people with Parkinson's disease develop salient hypokinetic dysarthria. However, it is unclear how extensive the voice impairment is in the non-advanced stages of Parkinson's disease. Therefore, the aim of the current research was to investigate the auditory-perceptual characteristics of voice in people with Parkinson's disease (PWPD) in non-advanced stages.

**Methods**: 29 PWPD and 32 healthy older controls were recruited. For each participant, a recording of the sentence *"We were away a year ago"* was acquired. These recordings were evaluated by two licensed and experienced speech-language pathologists, who provided perceptual ratings of overall dysphonia severity, breathiness, roughness, and perceived age.

**Results**: MANCOVA analysis showed that, when controlling for age and intensity, there was a significant effect of group (p=0.001) on perceptual voice quality. PWPD were perceived to be significantly older, more breathy and more severely dysphonic than the older healthy controls. No differences were found for the perceived roughness.

**Conclusions**: The results suggest that features of hypokinetic dysarthria in voice, specifically breathiness, are present in non-advanced stages of PWPD and may contribute to listener perceptions of speaker age. Moreover, the perceptual voice profiles in PWPD showed great variability, possibly reflecting the heterogeneity of disease impact on individuals. The results of this study may inform how research targets rehabilitation and maintenance of voice and laryngeal function in PWPD, specifically those at non-advanced stages.

**Keywords:** Parkinson's disease – non-advanced stages – hypokinetic dysarthria – voice – auditory-perceptual assessment

#### Introduction

Parkinson's disease (PD) is a neurodegenerative disorder that affects multiple systems and pathways (Sveinbjornsdottir, 2016). People with PD (PWPD) exhibit both motor symptoms, such as bradykinesia, rigidity, and tremor, and nonmotor symptoms, such as apathy, sleep problems, and memory complaints (Sveinbjornsdottir, 2016). One of the suggested mechanisms relating to etiology and disease severity in PD is age. Levy (2007) states that age is related to the disease severity in PD more so than duration. On the other hand, there have been mentions of PD as "accelerated aging" (Levy, 2007). More recently, Rodriguez et al (2015) explored the possibility of PD as a result of aging. They found that the neurodegeneration in PD occurs in brain areas that also typically degenerate during a healthy aging process (Rodriguez et al., 2015). Rodriguez et al (2015) concluded that the etiological factors of PD are multifactorial, which can explain the variability seen in the disease. However, they also suggested that many etiological factors involved in PD are the same as the factors involved in aging.

The motor and nonmotor impairments affecting PWPD are heterogeneous, especially in the nonadvanced stages of the disease (Thenganatt & Jankovic, 2014; von Coelln & Shulman, 2016; Wolters, 2008). However, the profile of voice impairment in PWPD is largely based on reports from populations who manifest clinically salient levels of dysarthria resulting in "hypophonia", which is perceived as low intensity and breathy voice quality (Miller, 2017; Stathopoulos et al., 2014). While the perceptual characteristics of voice impairment in hypophonic PWPD are well understood, the prevalence of voice impairment in those PWPD at different stages of disease progression is less clear. For example, the speech changes that occur prior to clinically salient hypokinetic dysarthria PD are not well

documented (Magee et al., 2019). Often, hypokinetic dysarthria has been proposed as an early marker of PD (Sapir, 2014; Stewart et al., 1995). For example, Orozco-Arroyave et al (2016) found that it was possible to distinguish PWPD and healthy older adults using analysis of continuous speech, even in the early- to middle stages, as did Ho et al (1999). In addition, measures of vowel articulation were significantly impacted in PWPD, even before dysarthria was perceived (Jan Rusz, Cmejla, et al., 2013).

When focusing specifically on voice characteristics, initial vocal changes may be subtle (Defazio et al., 2016; Lirani-Silva et al., 2015; Miller, 2017; Jan Rusz, Čmejla, et al., 2013; Sapir, 2014). However, voice has been noted as an early sign of PD that can be affected more than other aspects of speech (Ho et al., 1999; Ma et al., 2020; Ramig et al., 2004). Rusz et al (2011) reported voice and speech impairments as measured by acoustic voice analysis in people with early untreated PD. Defazio et al (2016) found that voice impairments may be more prevalent than articulation disorders in the early stages of PD, but that they may not yet result in perceived disability. Moreover, Lirani-Silva et al (2015) found that the acoustic measures of voice differed between PWPD in nonadvanced stages and healthy older adults, but did not find that difference for perceptual measures. On the other hand, Holmes et al (2000) found both acoustic and perceptual differences, where perceptual voice quality of PWPD showed reduced variability and loudness and increased harshness and breathiness (Holmes et al., 2000). These studies suggest that, while the voice impairments in PWPD may already be measurable by acoustic analyses in nonadvanced stages of the disease, there is no consensus in the literature if changes in vocal function can be perceived in nonadvanced stages of PD. Moreover, it is unclear if the voice impairments that might be present in nonadvanced stages also reflect a vocal disability.

Another challenge to describing the voice of PWPD in nonadvanced stages is that the perceptual characteristics of voice in PWPD present very similar to the perceptual characteristics of aging. The hallmark perceptual features of presbyphonia are among other things a reduction in loudness, an increase in breathiness, hoarseness and vocal instability (Galluzzi & Garavello, 2018; Ramig et al., 2001). Similarly, hypokinetic dysarthria is characterized by a voice that is more quiet, hoarse and breathy (Brabenec et al., 2017; Lirani-Silva et al., 2015). As Rodriguez et al (2015) suggested that aging and PD may have similar etiologies, it would be logical to consider that the voice disorders associated with PD and aging present in a similar pattern. Therefore, it is possible that the voice changes in PWPD are not distinguishable from the voice changes related to aging especially in the nonadvanced stages of the disease. Lirani-Silva et al (2015) indeed found that, while there are significant differences in acoustic parameters of voice.

The inception of laryngeal impairments in PD signals the initiation of a process during which voice function will decline over a period of years, even when motor complaints remain fairly stable (Holmes et al., 2000; Magee et al., 2019; Skodda et al., 2013). As PWPD reach stages where voice impairments affect activities and participation to a self-perceived critical level, they will often seek services of the speech-language pathologist. However, at any stage of the disease, interventions should aim for the best possible level of communication (Miller, 2017). Given that there are changes in speech and voice in the early stages of the disease (Magee et al., 2019; Stewart et al., 1995), it may be beneficial to start intervention as soon as possible after diagnosis. However, there is little evidence for the effectiveness of voice therapy early on in the disease process (Ciucci et al., 2013). Therefore,

understanding how voice impairment manifests at different time-points, and specifically in the nonadvanced stages, is critical for advancing our knowledge of vocal function decline in PD. In addition, this knowledge is needed to inform the development of new treatments which may hold potential to moderate motor decline and/or maintain current functional levels, and thus the quality of life, for extended periods. The purpose of the current study was to investigate auditory-perceptual characteristics of voice in PWPD at nonadvanced stages. Our hypotheses were that the auditory-perceptual measures of vocal function would be similar for a group of speakers with PD at nonadvanced stages of the disease when compared to a group of older speakers without PD within a similar age range.

## Methodology

**Participants:** This study was approved by a university Institutional Review Board (IRB # 1709-026-1809-CR2). Sixty-one male adult speakers were recruited into 2 different groups: PWPD (n = 29), older healthy controls (n = 32). The PWPD were in nonadvanced stages of PD, defined in this study as Hoehn & Yahr (HY) levels 1 to 3. The final determination of the HY level was made by consensus of the 1st and 2nd authors and was based on assessment at the time of participation in the study. Additional inclusion criteria for participants in the PWPD group included a diagnosis of PD by a neurologist and no other history of comorbid neurological disease. Inclusion criteria for healthy older adults consisted of age at least 50 years or above and no history of diagnosed neurological disease, or vocal complaints. All participants were recruited via convenience sampling from the local community.

**Instrumentation and Procedures**: To perform auditory-perceptual assessments, speech recordings were first acquired using the Computerized Speech Lab (CSL – Pentax

Medical, Montvale, NJ) and a head-mounted condenser microphone (model C520; AKG Acoustics, Northridge, CA) placed approximately 3cm from the left corner of a participant's mouth. All recordings took place in a laboratory setting on a university campus. The laboratory consisted of a sound-treated room with less than 45 dB background noise. Participants were asked to produce the sentence "We were away a year ago" from the CAPE-V protocol (Kempster et al., 2009) at a self-selected comfortable pitch and loudness. All recordings were digitized at a 44kHz sampling rate and saved as a sound file.

Two licensed and certified speech-language pathologists each with more than 5 years of experience in assessment and treatment of dysphonic speakers completed the auditoryperceptual judgments of speaker age and dysphonia severity. The CAPE-V scale (a visual analog scale ranging from 0 to 100, where 0 = no deviation from normal expectations and 100 = maximum deviation from normal expectations) was used to measure ratings of perceived dysphonia severity, roughness, and breathiness. Raters were also asked to judge the perceived age of each speaker. The raters were asked to focus on only the voice quality while rating the voice samples and they were blinded for the group membership of each speaker.

Sixty-one sound files were created, each consisting of 2 repetitions of the same sentence with a silent pause of 2 seconds in between for each speaker. Additional sound files serving as perceptual anchors for age, dysphonia severity, roughness, and breathiness were selected by the investigators. These anchors were utilized for a perceptual calibration task prior to any ratings of speakers. Sentence recordings of a 20-year-old speaker, a 70-year-old speaker, and speakers with mild and severe dysphonia, breathiness and roughness, respectively, were played for the raters with descriptions by the investigators. All raters were provided with the same perceptual anchors before starting the evaluation of the samples.

Once the raters understood the task and indicated perceptual calibration of speaker age and voice quality, the rating task commenced. The samples were rated in a sound-treated room with less than 45 dB background noise. The raters were asked to indicate a comfortable loudness to play the samples. All samples were then played at that comfortable loudness level. Sound files were randomized and then played twice to each rater. The same randomization of files was used for both raters. After the rater listened to the 2 repetitions of a speaker, they rated the 4 perceived dimensions (age, dysphonia severity, roughness, breathiness). All audio files were played twice, in random order, to allow for assessment of perceptual intra-rater reliability.

**Analyses**: The statistical analysis was performed in SPSS (IBM Corp. Released 2017. IBM SPSS Statistics for Windows, Version 25.0. Armonk, NY: IBM Corp.). Prior to analyses, outliers in each data set were removed based on the criteria of the SPSS program (data points falling outside 1.5x interquartile range). Descriptive statistical analyses were performed, after which the different groups were compared using parametric statistics. Because participants were not expressly matched for age, chronological age was considered as a covariate in the subsequent statistical analysis. In addition, vocal intensity as a measure of sound pressure level (SPL) was used as a covariate in the analyses. A MANCOVA was applied to the data sets, with participant group (PWPD and older healthy adults) as a between-subjects factor, the 4 variables (perceived age, dysphonia severity, roughness, and breathiness) as within-subject factors, and chronological age and vocal intensity as

covariates. The inter- and intra-rater reliability was calculated using intraclass correlation coefficients. The mean score of all raters was used for all statistical analyses.

#### Results

The mean chronological age of the PWPD was 68.76 years (SD = 8.10 years), while the mean chronological age of the older healthy participants was 66.28 years (SD = 8.54 years). An independent samples t test showed that both groups were not statistically different for chronological age (t[59] = -1.16, P = 0.251) or for loudness (t[59] = -1.44, P = 0.15). Descriptive data for each dependent variable across each participant group is reported in Table 4 and the data trends are illustrated in Figures 4 through 7. There was a clear distinction in the mean perceived age between the 2 groups, characterized by a 7 year perceived difference in PWPD and older healthy participants, which was noticeably different from the chronological age difference. The perceptual variables of overall dysphonia severity and breathiness were numerically greater in the PWPD group compared to the control group, with exception to roughness. Moreover, the spread was noticeably bigger in the PWPD compared to the older healthy adults, as indicated by larger standard deviations and interquartile ranges.

Prior to analysis, 1 outlying data point was removed from the data set of perceived breathiness, and 2 outlying data points were removed from the perceived severity and roughness. Results from the MANCOVA revealed significant differences between the groups (F[4, 51] = 5.59, Pillai's trace = 0.31, P = 0.001, partial  $\eta^2 = 0.31$ ). Follow-up analysis revealed that PWPD were perceived as significantly older than the healthy older adults (F[1]=7.242, P = 0.009, partial  $\eta^2 = 0.12$ ), as well as significantly more severely dysphonic (F[1] = 17.084, P < 0.001, partial  $\eta^2 = 0.24$ ) and significantly more breathy (F[1] = 19.57, P

< 0.001, partial  $\eta^2 = 0.27$ ). However, there was no significant difference for perceived roughness between the 2 groups (F[1] = 0.32, *P* = 0.57, partial  $\eta^2 = 0.006$ ).

# Table 4

Means and Standard Deviations for Perceptual Variables in Each Participant Group.

Variable name		Older Healthy Participants (n=32)	Participants with PD (n=28)
Chronological	Mean (SD)	66.28 (8.54)	68.76 (8.10)
Age	Median (IQR)	67.00 (58.25-75.00)	68.00 (64.50-76.00)
H&Y	H&Y stage 1	n/a	1
	H&Y stage 2	n/a	10
	H&Y stage 3	n/a	18
Time since PD	Mean (SD)	n/a	4y1m (3y3m)
diagnosis	Median (IQR)	n/a	2y11m (2y1m-5y11m)
Average	Mean (SD)	70.24 (3.00)	71.44 (3.50)
intensity (dB)	Median (IQR)	70.40 (67.58-72.70)	72.47 (68.96-73.99)
Perceived age	Mean (SD)	44.80 (9.18)	51.68 (12.11)
	Median (IQR)	43.50 (38.56-52.25)	51.75 (42.75-63.13)
Perceived	Mean (SD)	19.37 (9.37)	31.29 (14.549)
Dysphonia	Median (IQR)	19.50 (13.06-26.13)	33.25 (18.75-44.50)
Severity			
Perceived	Mean (SD)	19.90 (11.08)	21.78 (13.91)
Roughness	Median (IQR)	19.50 (10.19-26.81)	21.50 (12.50-29.00)
Perceived	Mean (SD)	7.27 (8.56)	23.59 (19.14)
Breathiness	Median (IQR)	3.00 (0.00-14.00)	18.50 (4.25-45.25)

Note. Perceived age is reported in years. Perceived dysphonia severity, roughness, and

breathiness are based on a scale of 0 (no deviation from normal) to 100 (maximum deviation

from normal).

Abbreviations: PD, Parkinson's Disease,

The reliability of the raters in this study was assessed using intra-class correlation coefficients (ICC). A 2 way random, consistency, single measures intra-class correlation was used for the inter-rater reliability, while a 2 way mixed, consistency, single measures intra-class correlation was used for the intra-rater reliability. All of the samples were rated twice to allow intra-rater reliability to be calculated. Cicchetti et al's (1994) guidelines were used to interpret the results. The first SLP had an overall excellent intra-rater reliability (ICC = 0.83-0.90, P < 0.001). The second SLP had an overall good intra-rater reliability (ICC = 0.73, 95%CI = 0.66-0.78, P < 0.001). Moreover, the overall interrater reliability was good (ICC = 0.60, 95%CI = 0.51-0.67, P < 0.001).

## Figure 4

Boxplots of the Perceived Age of Both Groups.



*Note.* The upper line of the colored box represents the third quartile (percentile 75), the middle line represents the median, while the lower line represents the first quartile (percentile 25). The ends of the whiskers designate the minimum and maximum values within 1.5x the interquartile range.



Boxplots of the Perceived Dysphonia Severity among Both Groups.

*Note.* The upper line of the colored box represents the third quartile (percentile 75), the middle line represents the median, while the lower line represents the first quartile (percentile 25). The ends of the whiskers designate the minimum and maximum values within 1.5x the interquartile range.



Boxplots of the Perceived Roughness among Both Groups.

*Note.* The upper line of the colored box represents the third quartile (percentile 75), the middle line represents the median, while the lower line represents the first quartile (percentile 25). The ends of the whiskers designate the minimum and maximum values within 1.5x the interquartile range.



Boxplots of the Perceived Breathiness among Both Groups.

*Note.* The upper line of the colored box represents the third quartile (percentile 75), the middle line represents the median, while the lower line represents the first quartile (percentile 25). The ends of the whiskers designate the minimum and maximum values within 1.5x the interquartile range.

## Discussion

The purpose of the current study was to explore the auditory-perceptual voice quality of PWPD in the early disease stages and to compare them with older healthy speakers without PD. In order to do so, 2 experienced speech-language pathologists rated samples of speakers representing these 2 groups. The comparison of these ratings revealed that the voice of PWPD was perceived to be significantly older, more breathy and more severely dysphonic than the voice of the older healthy adults. The results of this study revealed that PWPD, even
in the nonadvanced stages, are already perceived as more breathy and with greater dysphonic severity than older healthy controls. These results are in accordance with what Ho et al (1999) and Holmes et al (2000) found: voice is already affected early on in the disease process of PWPD. However, interestingly, Lirani-Silva et al (2015) did not find theseperceptual differences in people with nonadvanced PD. On the other hand, when looking at the intensity values of the speech samples, the intensity of the PWPD and healthy older controls did not differ significantly. This could suggest that intensity is not yet affected in the nonadvanced stages of PD. While breathiness and low intensity are the hallmark characteristics of hypokinetic dysarthria (Gracco et al., 1992; Holmes et al., 2000), it is unclear what factors predict the emergence of these perceptual features in PWPD, other than the presence of the disease. There is insufficient literature available to provide a clear understanding of the voice characteristics in early-stage PD, and additional studies that build on those from this investigation are needed to inform our knowledge.

Ma et al (2020) summarized the physiological correlates of acoustical changes in voice of PWPD. A common finding in PWPD was incomplete glottal closure caused by insufficient vocal fold adduction or bowing (Ma et al., 2020). This incomplete closure then results in an air leak, which could explain the breathiness we found in the current study. However, this incomplete closure may not have impacted subglottal pressure enough to influence sound pressure, which could be why there was no difference in vocal intensity for the PWPD and the older healthy participants in the current study. Moreover, Ma et al (2020) found that the vocal folds in PWPD vibrated more asymmetrically. Along with breathiness, this phenomenon may help to explain the perceived severity of dysphonia found in our study. Sapir (2014) further explains the underlying mechanisms to some of these vocal changes.

Due to the damage to the basal ganglia, PWPD may underscale their motor movements and have issues with sensory processing (Sapir, 2014), thus not maintaining the adequate vocal effort for good vocal quality.

Throughout the disease process, PD expresses itself variably (Wolters, 2008). PD comprises at least 2 clinically different variants: an akinetic (ie, nontremor dominant) and a tremulous form (ie, tremor-dominant) (Wolters, 2008). Moreover, there is variability in both the motor and nonmotor expression of the disease (Wolters, 2008). Similarly, the hypokinetic dysarthria associated with PD may present itself in variable ways (Sapir, 2014). There is no straightforward relationship between disease severity and the extent of dysarthria (Majdinasab et al., 2016; Metter & Hanson, 1986). However, the literature on this topic is not conclusive as Dias et al (2016) found a relationship between disease severity and speech impairment. In our current sample, the variability for the PWPD was notably larger than those for the healthy older adults. This suggested that there was overall greater variability in how the voices of PWPD were perceived compared to how the voices of the healthy older participants were perceived.

When looking at the perceptual assessment of age, it is worth noting that the older healthy participants and the PWPD were perceived as younger than their chronological age. It has been previously established that age can be assessed perceptually (Goy et al., 2016; Shipp & Hollien, 1969). Listeners distinguish young voices from older voices based on their perceptual characteristics, even when those elderly voices are not pathological. Elderly voices contain more tremor and voice breaks and are perceived as more hoarse and lower in pitch, as well as slightly breathy, weaker and strained (Galluzzi & Garavello, 2018; Gorham-Rowan & Laures-Gore, 2006; Mueller, 1997; Ramig et al., 2001). These voice differences

can be attributed to a multitude of organic changes: aging changes the histology of the vocal folds and affects the laryngeal muscles (Galluzzi & Garavello, 2018). Moreover, neuromuscular changes and calcification of the laryngeal cartilages also play a role in the changed sound (Galluzzi & Garavello, 2018). However, Goy et al (2016) also found that the age of older speakers typically is underestimated. They suggested that this could be caused by a limited presence of acoustic features to distinguish older from younger speakers (Goy et al., 2016). This may have been the case in the current speaker sample as well: as only healthy older adults without vocal complaints were included, they likely did not show salient levels of presbyphonia, and therefore would be perceived as younger. On the other hand, while the PWPD were perceived as younger than their chronological age, they were also perceived to be significantly older than the older healthy controls, even after controlling for chronological age of all participants. As perceived breathiness is one of the characteristics of the aging voice, and the PWPD and older healthy controls differed on this parameter, it is possible that breathiness caused the raters to judge the PWPD as older compared to the older and younger healthy controls. However, in the current study, we did not investigate other perceptual acoustic parameters often considered in older voices. Therefore, the groups may have differed as a result of parameters that were not assessed in this study.

The voice characteristics of speakers in the nonadvanced stages of PD are relevant to clinical practice. Up to 90% of the PWPD will experience communication difficulties, however, less than half of the PWPD will be seen by a speech-language pathologist (Nijkrake et al., 2009; Schalling et al., 2017). The majority of the PWPD who do see a speech-language pathologist receive assessment and treatment specifically for voice disorders (Schalling et al., 2017). The literature suggests that referrals of PWPD to speech-language pathologists occurs

in the later stages of the disease when the symptoms become prominent and disabling (Domingos et al., 2013). In earlier stages, the communication disorders may be too subtle compared to other symptoms of PD to warrant treatment. For example, Schalling et al (2017) found that most of the PWPD who did not receive speech therapy indicated there was no need for it. However, in later stages of PD, the speech and voice impairments may be more difficult to treat due to severity (Ramig et al., 2001). The current study adds to the extant literature in providing evidence that voice disturbances in PWPD are already present in the nonadvanced of the disease. It may, therefore, be of value to PWPD to initiate exercise-based voice treatment programs earlier than current clinical practice prescribes. Although studies investigating the prophylactic application of voice exercise treatments in PWPD are lacking (Ciucci et al., 2013), the vast literature on voice exercise programs for PWPD along with 2 recent systematic reviews provide strong evidence that targeted exercise programs can improve muscle function in PWPD and improve quality of life associated with balance, mobility, and communication (Alves Da Rocha et al., 2015; Ramig et al., 2008; Roeder et al., 2015).

## Limitations

There are a number of methodological factors that should be taken into account when considering the generalization of results from this study. As an initial study in a new line of investigation, the sample size was small considering the worldwide population of PWPD, and no a priori power testing was performed There was a lack of gender diversity, as only male voices were considered. Only perceptual measures were studied in this investigation, and no acoustic measures were included as variables. Future research may include more variables to develop a multidimensional profile of voice impairment in PWPD at nonadvanced stages.

Another limitation is that vocal intensity was not controlled for completely. While intensity was controlled for as a covariate in the statistical analyses, all participants were instructed to produce sentences at their self-perceived "comfortable loudness and pitch". Moreover, the raters were allowed to play the samples back on loudness comfortable for them. Future research should take these potential methodological concerns into account. Furthermore, only a single repetition of a sentence was used. PWPD's speech is variable in the nonadvanced stages of the disease (Thenganatt & Jankovic, 2014; von Coelln & Shulman, 2016; Wolters, 2008). It, therefore, stands to reason our samples may not have been representative of the variability in voice quality of PWPD at a specific point in time (the time of recording). In addition, Hoehn & Yahr staging was not consistently reported in medical records from community neurologists. All staging was conducted by the authors, with confirmation by the 2nd author who has more than 20 years of experience in assessing PWPD. Finally, both the inter- and intra-rater reliability was fairly low, despite having trained both speech-language pathologists prior to the evaluation task. Therefore, the current data has to be interpreted with caution. In follow-up research, more extensive training should be considered.

### Conclusion

The aim of the current research was to determine the auditory-perceptual voice characteristics in early-stage PWPD. The voice of PWPD was perceived as more severely dysphonic, more breathy, and older than the voice of healthy controls. This suggests that voice problems in PD are apparent in early, nonadvanced stages of the disease process.

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# IV. Self-Perceived Affective, Behavioral, and Cognitive Reactions Associated with Voice and Communication in People with Parkinson's Disease

#### Abstract

Introduction: The hypokinetic dysarthria in people with Parkinson's disease (PWPD) leads not only to speech difficulty but also to a decrease in psychosocial wellbeing. PWPD experience negative feelings and thoughts due to their communication changes. They participate less in communication, actively avoid speaking, or use coping strategies. Communication partners also experience and can provide information on these psychosocial aspects of communication. The aim of this study was, therefore, to investigate the affective, behavioral, and cognitive reactions related to voice disorders in PWPD and their communication partners and to compare those with the reactions of healthy controls.

**Methods**: Participants were recruited between September 2020 and March 2021 via convenience and snowball sampling. The four Behavior Assessment Battery-Voice (BAB-Voice) subtests, the Behavior Checklist (BCL), Communication Attitude Test for Adults (BigCAT), Speech Situation Checklist - Emotional Reaction (SSC-ER), and the Speech Situation Checklist - Speech Disruption (SSC-SD) were administered to 31 PWPD and 19 healthy controls. Each participant's respective communication partner filled out an adapted version of the BAB-Voice. The instrument was filled out either clinician-guided or participant-guided. The overall subtest scores were calculated and analyzed.

**Results**: No significant effect of administration mode was found (clinician-guided vs. participant guided) (Pillai's Trace=0.088, F[4]=0.970, p=0.435,  $\eta_p^2$ =0.088). All BAB-Voice subtest scores were significantly higher in PWPD compared to healthy controls (Pillai's Trace=0.380, F[4]=6.116, p=0.001,  $\eta_p^2$ =0.380). The communication partners of PWPD rated

the speech-related attitude, coping behaviors, speech disruptions and negative emotion very similar to the PWPD themselves. Their ratings consequently did not different significantly (Pillai's trace = 0.247, F[4]=2.216, p=0.094,  $\eta_p^2$ =0.247). Moreover, the affective, cognitive and behavioral reactions experienced all correlated highly with one another in the current sample. Finally, similar to other BAB-research, the items within the different subtests showed excellent internal consistency.

**Conclusions**: The BAB-Voice can be useful to describe the psychosocial effects of hypophonia in PWPD as well as to inform better diagnostic conclusions and treatment planning for voice and other communication impairments in PWPD. Furthermore, this study supports the supposition that the perceptions of communication partners related to voice and speech difficulties are aligned with those of the speakers with PD. Thus, the communication partners of PWPD can serve as a valuable source of information to both the individual with PD and healthcare providers.

**Keywords:** Parkinson's disease – hypophonia – psychosocial effects – communication partners

### Introduction

Parkinson's disease (PD) is a neurodegenerative disease associated with degeneration in the basal ganglia and nigrostriatal pathways (Sveinbjornsdottir, 2016). The subsequent dopamine deficiency causes the typical motor symptoms of rigidity, bradykinesia, resting tremor, and postural instability (Bartels & Leenders, 2009; Jankovic, 2008; Sveinbjornsdottir, 2016). Up to 90% of people with Parkinson's Disease (PWPD) also experience a unique cluster of speech and voice impairments (Miller, 2017; Ramig et al., 2004), which are collectively called hypokinetic dysarthria. Hypokinetic dysarthria presents as speech with imprecise articulation, low vocal intensity with harsh and breathy voice quality, and disturbance of prosody, among other characteristics (Brabenec et al., 2017; Magee et al., 2019; Ramig et al., 2004). Specifically for the vocal subsystem, hypophonia is apparent, which is characterized by lowering of vocal intensity, changes to vocal pitch, less variable intonation, and breathy voice quality with or without a vocal tremor (Holmes et al., 2000; Magee et al., 2019; Miller, 2017; Sewall et al., 2006). The communication changes associated with hypokinetic dysarthria have a significant negative impact on the quality of life and participation of PWPD (Dashtipour et al., 2018).

Multiple studies have investigated the experience of PWPD regarding the speech changes they have experienced. Several qualitative studies identified that the communication difficulty in PD was more dynamic than solely hypokinetic dysarthria (Johansson et al., 2018; Whitehead, 2010; Yorkston et al., 2017). PWPD experienced negative feelings such as anxiety, stress, fatigue, frustration, and embarrassment related to their speech (Johansson et al., 2018; Whitehead, 2010; Yorkston et al., 2017). Miller et al. (2008) quantified these changes and found that these experiences were independent of motor and cognitive status and

disease duration. The speech difficulty hindered PWPD in maintaining relationships (Whitehead, 2010) or communicating with people they do not know because of the stranger's reactions to the disordered speech (Johansson et al., 2020; Yorkston et al., 2017). PWPD also presented with difficulty with intelligibility in specific situations, notably talking in noisy situations (e.g., pubs, restaurants, with the television on) or on the phone (Schalling et al., 2017; Whitehead, 2010; Yorkston et al., 2017). Overall, PWPD experienced a loss of participation because of their speech issues (Schalling et al., 2017). Due to the experienced fatigue and the perceived increase in effort needed to communicate, many of the PWPD reported that they became more withdrawn, avoided situations and people, or took less initiative in conversation (Johansson et al., 2018; Martin, 2015; Yorkston et al., 2017). This loss of participation has even been measured quantitatively. Gustafsson et al. (2019) measured voice use using a portable voice accumulator and found that PWPD use their voice 50-60% less than their healthy counterparts. Other coping behaviors described include preparing what to say, adjusting their speech, repeating information, or informing people about their PD-related difficulty (Johansson et al., 2020; Schalling et al., 2017; Whitehead, 2010).

While wide-ranging instruments can describe these psychosocial reactions to the communication disorders in PWPD, none of the existing instruments have provided a comprehensive description of the emotional, cognitive, and behavioral reactions that PWPD experience when they communicate. The Communicative Effectiveness Survey (Donovan et al., 2008; Hustad, 1999), validated for use in PWPD (Dykstra et al., 2015), contains 8 questions on the effectiveness of communication. Similarly, the Communication Participation Item Bank (Baylor et al., 2009; Yorkston et al., 2008), also calibrated to PWPD

(Baylor et al., 2013), focusses on one specific aspect of communication: participation. The Dysarthria Impact Profile (Walshe et al., 2009), which has been used in PWPD (Cardoso et al., 2018; Letanneux et al., 2013), collects information on the psychosocial concerns of PWPD but includes only limited situations and coping behaviors. In contrast, instruments used to record data on psychosocial characteristics associated with other communication disorders, such as stuttering, have thoroughly assessed these affective, behavioral, and cognitive reactions in multiple speech situations (Vanryckeghem et al., 2004, 2017; Vanryckeghem & Brutten, 2011, 2012, 2018; Wesierska et al., 2018). One example of such an instrument is the Behavior Assessment Battery (BAB, Vanryckeghem & Brutten, 2018). This tool was originally developed for people who stutter (PWS) and was adapted later to the Behavior Assessment Battery – Voice (BAB-Voice) for a population with the neurological voice disorder spasmodic dysphonia (SD; Vanryckeghem et al., 2016; Vanryckeghem & Ruddy, 2015; Watts & Vanryckeghem, 2017). The BAB-Voice incorporates four subtests to describe the presence of specific coping behaviors (Behavior Checklist, BCL), the way a speaker thinks about their voice (Communication Attitude Test for Adults, BigCAT), the negative emotion generated by specific speech situations (Speech Situation Checklist Emotional Reactions, SSC-ER), and the extent of vocal disturbance caused by specific speech situations (Speech Situation Checklist Speech Disruptions, SSC-SD) (Vanryckeghem et al., 2016; Vanryckeghem & Ruddy, 2015; Watts & Vanryckeghem, 2017).

Communication requires the active engagement of a partner. Consequently, close communication partners (i.e., the communication "proxy" or "proxies") of PWPD are a valuable source of information, and their opinions should be considered when evaluating the communication experiences of PWPD (Gillivan-Murphy et al., 2019b). PWPD have shown

issues with self-perception (Parveen & Goberman, 2017; Sapir, 2014), and thus their judgment of communication exchanges may be different from that of a communication partner. Previous research showed that communication partners tend to rate the psychosocial consequences of dysarthria, such as voice handicap, feelings, and communicative effectiveness, as less impaired than the PWPD themselves (Donovan et al., 2008; Miller et al., 2008; Parveen & Goberman, 2017). However, this finding is not ubiquitous in the research literature. As an example, Dykstra et al. (2015) did not find significant differences between proxy and PWPD ratings. More information on the association of proxy and speaker ratings is needed, as the perceptions and assistance of frequent communication partners may become more important once communication difficulties of the PWPD increase during the disease process. In addition to assessing the communication experience of a speaker, the BAB-tool has previously been adapted to allow for the assessment of the affective, behavioral, and cognitive reactions of speakers by their life partners (Svenning et al., 2021). When evaluating life-partners and PWS, ratings of communication experiences were found to be similar. Therefore, the BAB-Voice was considered an appropriate tool to adapt to communication partners of PWPD.

Considering and measuring the above-described affective, cognitive, and behavioral reactions to hypophonia is important for several reasons. Typical voice therapy in PWPD is unidimensional and does not diagnose or treat the psychosocial consequences of the speech and voice disorders in PWPD (Gillivan-Murphy et al., 2019b; Yorkston et al., 2017). Rather, it typically focuses on the voice handicap itself, such as aspects of voice quality, vocal stamina, or voice skills needed for professional and/or societal functions (Gillivan-Murphy et al., 2019b). Even post-treatment, negative affective reactions may not be resolved (Gillivan-

Murphy et al., 2019b; Spurgeon et al., 2015). Therefore, the purpose of this study was to investigate the affective, behavioral, and cognitive reactions related to voice disorders in PWPD as perceived by themselves and by communication partners respectively and to compare with healthy older counterparts. Based on existing reports, the hypothesis was that the emotional reactions, attitude, and coping behaviors related to voice use would be different in PWPD compared to healthy adults. Moreover, it was hypothesized that the responses of close communication partners on the same dimensions would be different than those of the PWPD. Given that this project was the first to administer the tool to PWPD, the relationship between the different reactions was investigated as well as the internal consistency of the instrument.

## Methodology

The protocol was approved by the Texas Christian University Institutional Review Board.

## Sample and Recruiting

Dyads consisting of adults with PD and their proxy (daily communication partner) were recruited. To participate in the study, PWPD had to (1) have a diagnosis of PD by a neurologist, (2) have a negative history of speech or voice problems prior to PD diagnosis, (3) have no other diagnosed neurological impairments unrelated to PD, and (4) have selfreported hearing within normal limits for their age with or without corrective amplification. Finally, they had to (5) have a common communication partner (e.g. partner, child, or caregiver, also referred to as "proxy" in this study) willing to participate in the study. "Common communication partner" was defined as one who interacts verbally with the PWPD participant daily. Proxy participants must (1) live with the PWPD or communicate

with the individual daily and (2) have self-reported hearing within normal limits for their age with or without corrective amplification. Pairs of healthy older participants (without PD) and a common communication partner were also recruited. Healthy controls had to (1) have no diagnosed neurological impairments, (2) have a negative history of speech or voice problems, (3) have self-reported hearing within normal limits for their age with or without corrective amplification, (4) be 50 years or older. For their communication partners, the same inclusion criteria as for the communication partners of PWPD were used.

To maximize the participant sample, two recruitment strategies were employed. One group of participants was recruited in pairs within the Dallas-Fort Worth metroplex (DFW), i.e. in the cities and towns in the area of Dallas, Fort Worth, and Arlington (sample A, see Data Collection Protocol). For sample A, a clinician-guided administration of the BAB-Voice was employed. An established volunteer database was used to contact PWPD and their communication partners. PWPD were contacted through either telephone or e-mail, based on their previous interest in research. Snowball sampling was also employed by asking the participants to share the information about the study with other interested persons. The healthy controls and their communication partner were recruited from the same area using social media, convenience, and snowball sampling.

A second sample was recruited from populations of PWPD outside of the DFW area and a participant-guided administration of the BAB-Voice was employed (sample B, see Data Collection Protocol) and. Invitations to participate in the research, as well as a link to the survey, were distributed online or via postal mail using PD support groups and social media. Prospective participants and their communication partners were asked to contact the

researchers through e-mail with questions regarding participating. Controls were recruited through social media and convenience sampling in the USA but outside of DFW as well.

An a priori power analysis was performed using G\*Power 3.1 (Faul et al., 2009) to determine the sample size for the main research question assessing a potential difference between disordered speakers and healthy controls. The analysis was based on Vanryckeghem et al.'s (2016) study, which used a version of the BAB-Voice in a population of people with SD and healthy controls. They found effect sizes between d=1.047 and d=3.4016. Based on these results, the sample size calculation for the global effect of a MANOVA was performed using the lowest effect size (d=1.047), an alpha level of p=0.05, and power of 0.80. The analysis included a single between-group factor (2 groups) over 4 different outcome measures. Prior to running it, Cohen's d was converted to  $f^2$  (=0.2735). The power analysis revealed a required total needed sample size of 50 to find a difference between disordered speakers and healthy controls.

#### Instrumentation

Instrumentation for PWPD and Healthy Controls. <u>Demographic data</u> were collected first and included: gender, age, country of origin, native language, state of residence, highest degree attained, current profession, diagnosis with PD (if PD, then onset month and year of PD, history of PD treatment, member of a PD support group or not), selfreported weekly exercise frequency, other neurological diagnoses, self-reported hearing status, and history of speech, voice, or language therapy (see Appendix 1).

Secondly, the participants completed the BAB-Voice assessment using an adapted version of that used by Watts and Vanryckeghem (2017). The following adaptions were made:

- Firstly, an explanation of "phonation" was provided prior to the first subtest and a summary of this explanation appeared prior to each subsequent subtest of the BAB-Voice.
- Any ambiguity regarding speech vs. voice was clarified within the questions of the instrument.
- Examples given within the BAB-Voice of disordered voice behaviors were tailored to the specific voice issues of PWPD.
- Finally, some situations in the SSC subtests were adapted to fit better with an older, possibly retired population, as is common in PWPD.

The BAB-Voice consisted of four subtests. The BCL looked at the coping behaviors. The participant indicated for 34 coping behaviors if they utilized them or not ("yes"-1 and "no"-0), resulting in a score between 0 and 34, with higher scores indicating more coping hehaviors. Additionally, the participant could indicate the frequency of the used coping behaviors, which was not part of the test score. Some examples of BCL items were "avoiding eye contact", "taking a deep breath before speaking", or "clearing your throat". The BigCAT contained 34 statements concerning the participant's voice-related attitude. For example, "There is something wrong with my voice", "My voice limits my future." The participant rated the statements as "True" or "False." If the participant's response reflected a negative attitude on voice, the item was scored as 1, whereas positive responses were scored as 0. This resulted in a possible score on the BigCAT between 0 and 34 points, with higher scores indicating more negative attitude on voice. The SSC-ER required the participant to rate the amount of negative emotional reaction on a 5-point Likert scale (1-"Not at all" to 5-"Very much") in specific speech situations, such as "talking on the phone", "talking to a stranger", "saying a sound or word that previously had been troublesome," etc. Thirty-eight speech situations were rated, resulting in a score ranging between 38 and 190, with higher scores indicating more negative emotional reaction to speech situations. Finally, the SSC-SD was organized similarly but the participant had to consider voice disruptions present in speech situations using the same Likert scale (1-"Not at all" to 5-"Very much"). The same 38 speech situations were provided, leading again to a possible score between 38 and 190. Higher scores indicated more experienced voice difficulty in different speech situations.

Instrumentation for Communication Partners. Data collection for the communication partners started with adapted demographic questions similar to those presented to the PWPD. The first part consisted of demographic questions asking about the communication partner's gender, age, country of origin, native language, state of residence, highest attained degree, and current profession. The questionnaire also asked if their communication partner was diagnosed with PD or not, how long they knew their communication partner, how often they talked with them, and if they themselves required hearing aids (see Appendix 2). After, the proxy filled out an adapted version of the BAB-Voice. All subparts of the instrument were adapted to make sure that the communication partner replied using the PWPD/control's perspective (e.g., "Is your communication partner anxious...", "Does your communication partner think..."). The subtests and their scoring remained the same. The assessments for the communication partners were integrated into the same Qualtrics as the BAB-Voice and demographic questions for the PWPD to allow for paired data analysis.

### **Data Collection Protocol**

**Sample A.** The clinician-guided sample was invited to schedule a time and date for a video call. During the video call, the clinician went over all questions with the participant, which allowed them to provide assistance and direction as needed. The participants were also able to see the questions in written form through screen sharing. The administration of the four BAB subtests was randomized for each participant. The PWPD and proxy filled out the test separately, without the other person listening in.

**Sample B.** The participant-guided sample had access to the same online version of the BAB-Voice shared with the clinician-guided sample. They completed the BAB-Voice independently via a self-guided mode. Participants were instructed to contact the investigator by e-mail with any questions or when needing clarification. Participants completed screening questions and indicated what group they belonged to (PWPD/healthy control – communication partner). They were then were directed to the correct version of the test if they were eligible to participate. The instructions encouraged the participant and communication partner to fill out their version of the BAB-Voice separately. After the demographic questions, the BAB-Voice-subtests were administered at random. As soon as the first half of the dyad finished their version of the BAB-Voice, their communication partner (or PWPD/control, depending on who started) was invited to complete the BAB-Voice.

#### Statistical Analysis

The main research questions of this project were: what affective, behavioral, and cognitive reactions related to vocal function do PWPD experience compared to healthy controls? What affective, behavioral, and cognitive reactions related to vocal function do

communication partners indicate in PWPD compared to healthy controls? Two betweensubjects factors were included: disease status (PD – control), administration mode (clinicianguided – self-guided), and one within-subjects factor: rating type (self – partner). The main dependent variables were coping behaviors (overall score on BCL), voice-related attitude (overall score on BigCAT), negative emotional reaction (overall score on SSC-ER), and voice disruption (overall score on SSC-SD).

Prior to all analyses, the overall scores for the different subtests were calculated by adding all item scores (see Instrumentation for scoring). In accordance with the manual, missing items were coded as 0 for the BCL and BigCAT and as 1 for the SSC-ER and SSC-SD to allow for the calculation of the total subscores. After, the data were visualized and analyzed descriptively. The demographic data were presented in a table with the mean, standard deviation, median, and interquartile range. The PD group was compared for these variables with the control group using Mann-Whitney U tests for the continuous variables (e.g. age) and chi-square tests for categorical variables (e.g. PD treatment). Similarly, demographic differences between the groups with different administration modes were determined. A significance level of  $\alpha$ =0.05 was adopted unless otherwise indicated. The overall scores of the subtests were presented across the groups with mean, standard deviation, median and interquartile range. Outliers outside 3x the interquartile range were removed prior to the inferential analyses.

To compare administration modes (participant-guides vs. clinician-guided), as well as disease status groups (PWPD vs. healthy adults), a two-way MANCOVA was used with chronological age as a covariate. Administration mode and disease status were used as the independent variables, and the subtest scores were the dependent variables. Follow-up analyses with Bonferroni correction were performed if needed. To compare the scores of PWPD with their communication partners', a two-way mixed, absolute agreement, single measures Intra Class Correlation (ICC) was employed to assess the inter-rater reliability, and a repeated-measures MANOVA was used to determine differences in the ratings. Rating type (self – partner) was a within-subject independent variable, with the subtest scores as dependent variables. Paired t-tests with Bonferroni correction were used for follow-up analysis if needed.

To assess the strength of association between the four BAB-Voice subtests, correlation analysis was performed. In pairs and with Pearson's correlation, the relationship between coping behaviors, speech-related attitude, emotional reactions, and vocal symptoms as described by the subtests (BCL, BigCAT, SSC-ER, SSC-SD) was determined. Finally, the internal consistency of the BAB-Voice was considered, as this instrument had not been used in this specific population yet. Two-way mixed, absolute agreement, average measures ICC were used to determine the internal consistency of each of the subtests.

#### Results

#### **Descriptive Statistics**

The final sample consisted of 50 dyads of participants (31 PWPD, 19 controls) with a daily communication partner. The overall characteristics of the sample can be found in Table 5. The clinician-guided sample was comprised of 16 PWPD and their communication partner as well as 10 controls and their communication partner. Similarly, the participant-guided sample included 15 PWPD and their communication partner with 9 controls and their communication partner. Overall, there were more males in the PWPD-group (58%), compared to the control group (36%), but this difference was not statistically significant

## Table 5

The Descriptive Data of	the Overall Sample of PWPD	and Healthy Controls
1 2	1 2	2

		PWPD (n=31)	Control (n=19)	Test result
Ppt Gender	Male (%)	18 (58.1%)	7 (36.8%)	$\chi^{2}(1)=2.122,$
	Female (%)	13 (41.9%)	12 (63.2%)	<i>p</i> =0.145
Ppt Chronological	Mean (SD)	71.23 (9.09)	63.79 (7.04)	U=455.0,
Age (years)	Median (IQR)	73 (65-78)	62 (59-69)	<i>p</i> =0.001
CP Gender	Male (%)	12 (38.7%)	13 (68.4%)	$\chi^2(1)=4.160,$
	Female (%)	19 (61.3%)	6 (31.6%)	<i>p</i> =0.041
<b>CP Chronological</b>	Mean (SD)	69.74 (9.35)	64.47 (8.04)	U=416.5,
Age (year)	Median (IQR)	71 (66-76)	65 (59-70)	<i>p</i> =0.015
PD duration	Mean (SD)	8.02 (5.13)		
(years)	Median (IQR)	7.33 (4-10.75)		
PD treatment	Treated	31 (100%)		
	- Medication	- 30 (96.8%)		
	- DBS	- 6 (19.4%)		
	- Other	- 7 (22.6%)		
SLT in past	SLT in past	12 (38.7%)		
	SLT currently	2 (6.5%)		

*Note*. Categorical data are presented with the absolute and percent frequencies, along with the results of a Chi-square test to determine the difference between the PWPD and control sample. Continuous data are presented with mean, standard deviation, median, and interquartile range, along with the results of a Mann-Whiney U test to compare PWPD and controls.

Abbreviations. PWPD, people with Parkinson's Disease; Ppt, participant; CP,

communication partner; PD, Parkinson's disease; SLT, speech-language therapy; SD,

standard deviation; IQR, interquartile range.

( $\chi^2(1)=2.122$ , p=0.145). The average participant age was 71.23 years (SD=9.09 years) for PWPD and 63.79 years (SD=7.04 years) for controls, which was a statistically significant difference (U=455.0, p=0.001). The samples also significantly differed for communication partner age (U=416.5, p=0.015): in the PD sample, communication partners were on average 69.74 years old (SD=9.35 years) compared to 64.47 years (SD=8.04 years) in the sample of healthy controls. The PWPD had significantly more female communication partners, whereas the controls had more male communication partners ( $\chi^2(1)=4.160$ , p=0.041). The communication partners had known their participants for similar durations (U=341.50, p=0.347): 38.45 years (SD=17.94) in the PD sample and 35.59 years (SD=14.96 years) in the control sample.

## Table 6

		PWPD (n=31)		Control (n=19)	
		Participant	СР	Participant	СР
BCL	Mean (SD)	7.19 (5.84)	7.06 (6.33)	2.68 (3.99)	1.11 (1.84)
	Median (IQR)	7 (2-9)	7 (2-10)	1 (0-4)	0 (0-2)
BigCAT	Mean (SD)	16.32 (11.71)	13.74 (11.51)	2.29 (2.26)	1.37 (1.12)
	Median (IQR)	13 (6-27)	12 (2-24)	2 (1-4)	1 (0-2)
SSC-ER	Mean (SD)	82.97 (46.75)	72.55 (39.62)	48.89 (11.35)	42.50 (6.27)
	Median (IQR)	60 (50-124)	57 (44-84)	46 (38-55)	39.5 (38-46.75)
SSC-SD	Mean (SD)	77.32 (40.17)	74.42 (38.21)	45.68 (9.24)	42.79 (7.30)
	Median (IQR)	65 (48-96)	63 (44-93)	42 (39-51)	39 (38-46)
	( )	. ,	. ,	. ,	. ,

*Note.* The average and median scores for each subtest are represented for both groups.

*Abbreviations*. PWPD, people with Parkinson's Disease; CP, communication partner; BCL, Behavior Checklist; BigCAT, Communication Attitude Test for Adults; SSC-ER, Speech Situation Checklist – Emotional Reaction; SSC-SD, Speech-Situation Checklist – Speech Disruption, SD, standard deviation; IQR, interquartile range.

# Table 7

# The Scores of the Clinician-guided and Participant-guided Sample of PWPD and Healthy

		Sample A: Clinician-guided sample			Sample B: Participant-guided sample				
		PWPD (n=16)		Control (n=10)		PWPD (n=15)		Control (n=9)	
		Ppt	СР	Ppt	СР	Ppt	СР	Ppt	СР
BCL	Mean	5.81	7.5	1.80	0.9	8.67	6.6	3.67	1.38
	(SD)	(4.55)	(6.32)	(2.30)	(2.18)	(6.82)	(6.52)	(5.27)	(1.41)
	Median	5	7	1	0	9	6	1	1.5
	(IQR)	(2-7.75)	(2.25-	(0-3.25)	(0-1)	(2-14)	(2-10)	(0-8.5)	(0-2)
			10)						
BigCAT	Mean	12.69	11.63	2.11	0.70	20.20	16.00	2.5	2.11
	(SD)	(11.52)	(10.44)	(1.62)	(0.82)	(10.97)	(12.51)	(2.93)	(0.92)
	Median	9.5	9	1	0.5	21	14	2	2
	(IQR)	(2-24.5)	(2-	(1-4)	(0-1.25)	(11-33)	(3-30)	(0.25-	(1-3)
			20.25)					3.5)	
SSC-ER	Mean	68.25	70.56	48.90	39.56	98.67	74.67	48.89	45.44
	(SD)	(37.39)	(29.50)	(10.62)	(2.55)	(51.71)	(49.22)	(12.76)	(7.58)
	Median	52.50	59.50	45.5	39	75	57	46	42
	(IQR)	(45.25-	(46-93)	(40.25-	(38-40)	(57-	(38-84)	(38-	(38-
		65.75)		57.25)		145)		56.5)	52.5)
SSC-SD	Mean	62.13	66.00	45.20	41.00	93.53	83.40	46.22	44.78
	(SD)	(24.52)	(23.91)	(7.61)	(5.62)	(47.63)	(48.45)	(11.24)	(8.71)
	Median	58	59.5	43	38.5	75	68	42	40
	(IQR)	(43.25-	(48.5-	(38-	(38-	(60-	(44-	(39.5-	(38-52)
		66)	84)	51.75)	41.75)	145)	109)	48.5)	

*Note.* The average and median scores for each subtest are represented for all groups.

Abbreviations. PWPD, people with Parkinson's Disease; Ppt, participant; CP,

communication partner; BCL, Behavior Checklist; BigCAT, Communication Attitude Test for Adults; SSC-ER, Speech Situation Checklist – Emotional Reaction; SSC-SD, Speech-Situation Checklist – Speech Disruption, SD, standard deviation; IQR, interquartile range.

Subtest scores were calculated for both the overall sample as well as the clinicianguided and participant-guided samples. Outlying data points were removed prior to analysis. The average scores, standard deviations, median, and interquartile range for the scores on the BCL, BigCAT, SSC-ER, and SSC-SD of both samples together can be found in Table 6. The average score of PWPD on the BCL was 7.19 (SD=5.84) compared to 2.68 (SD=3.99) in controls, indicating a higher presence of coping behaviors in PWPD. PWPD also presented with more negative speech attitudes, indicated by higher scores on the BigCAT (mean=16.38, SD=11.71) compared to controls (mean=2.29, SD=2.26). In the different speech situations, PWPD also showed more experienced negative emotional reaction (mean=82.97, SD=46.75) and more experienced voice disruption (mean=77.32, SD=40.17) compared to the control population (SSC-ER mean=48.89, SD=11.35; SSC-SD mean=45.68, SD=9.24). Overall, the subscores of PWPD were numerically higher than those of the healthy adults (for the statistical comparison: see later). This statement held true when comparing the PWPD's and control's communication partners' ratings. The found differences in subscores for PWPD and controls were fairly small for some subtests (4 points on the participant BCL) or as big as approximately 30 points for other subtests (participant SSC-ER and SSC-SD).

The demographic characteristics were compared across administration mode of the samples (clinician-guided sample vs. participant-guided sample) as well. Chi-square tests revealed that both samples did not differ for the gender of the participant ( $\chi^2$ [1]=0.321, p=0.571) or the gender of the communication partner ( $\chi^2$ [1]=0.321, p=0.571). Similarly, no difference was found for participant age (U=278.00, p=0.509), communication partner age (U=333.50, p=0.676), or the duration of the acquaintance of the participant and communication (U=260.50, p=0.317). Specifically for the PWPD, the two groups did not

significantly differ for disease duration (U=92.00, p=0.281). The scores of the two samples on the BAB-Voice subtests are described in Table 7. When comparing both samples, the scores of the PWPD appeared to be numerically higher in the clinician-guided sample compared to the participant-guided sample for the BigCAT, SSC-ER, and SSC-SD. However, the participant-guided sample also presented with more variability. The other subtests and participants scores appeared to be similar across both samples. The statistical comparison is described later.

## Research Questions 1 and 2: The Effect of Administration Mode and Disease Status

To determine the difference in scores between the administration modes and the disease status of the participants, a two-way MANCOVA was employed to control for chronological age. There was no significant effect of the covariate age (Pillai's Trace=0.027, F[4]=0.276, p=0.891,  $\eta_p^2=0.027$ ), of the independent variable administration mode (Pillai's Trace=0.088, F[4]=0.970, p=0.435,  $\eta_p^2=0.088$ ), or of the interaction between administration mode and disease status (Pillai's Trace=0.102, F[4]=1.131, p=0.356,  $\eta_p^2=102$ ). There was, however, a significant effect of disease status on the BAB-Voice subtest scores (Pillai's Trace=0.380, F[4]=6.116, p=0.001,  $\eta_p^2=0.380$ ). The descriptive data as well as the between-subjects effects (a one-way ANOVA follow-up analysis) revealed that PWPD scored significantly higher than the healthy controls on all subtests: BCL (F[1]=10.145, p=0.003,  $\eta_p^2=0.191$ ), and SSC-SD (F[1]=11.47, p=0.002,  $\eta_p^2=0.211$ ) (see Figures 8-11).

Self-rated Scores of PWPD and Healthy Adults on BCL.



*Note*. The upper line of the colored box represents the third quartile (percentile 75), the middle line represents the median, while the lower line represents the first quartile (percentile 25). The ends of the whiskers designate the minimum and maximum values within 1.5x the interquartile range, while the circle represents outliers between 1.5 and 3x interquartile range. *Abbreviations*. BCL, Behavior Checklist; PWPD, people with Parkinson's disease.



Self-rated Scores of PWPD and Healthy Adults on BigCAT.

*Note*. The upper line of the colored box represents the third quartile (percentile 75), the middle line represents the median, while the lower line represents the first quartile (percentile 25). The ends of the whiskers designate the minimum and maximum values within 1.5x the interquartile range, while the circle represents outliers between 1.5 and 3x interquartile range. *Abbreviations*. BigCAT, Communication Attitude Test for Adults; PWPD, people with Parkinson's disease.



Self-rated Scores of PWPD and Healthy Adults on SSC-ER.

*Note*. The upper line of the colored box represents the third quartile (percentile 75), the middle line represents the median, while the lower line represents the first quartile (percentile 25). The ends of the whiskers designate the minimum and maximum values within 1.5x the interquartile range, while the circle represents outliers between 1.5 and 3x interquartile range. *Abbreviations*. SSC-ER, Speech Situation Checklist – Emotional Reaction; PWPD, people with Parkinson's disease.



Self-rated Scores of PWPD and Healthy Adults on SSC-SD.

*Note*. The upper line of the colored box represents the third quartile (percentile 75), the middle line represents the median, while the lower line represents the first quartile (percentile 25). The ends of the whiskers designate the minimum and maximum values within 1.5x the interquartile range, while the circle represents outliers between 1.5 and 3x interquartile range. *Abbreviations*. SSC-SD, Speech Situation Checklist – Speech Disruption; PWPD, people with Parkinson's disease.

#### **Research Question 3: Comparison between Participants and the Communication Partners**

The ICC between the ratings of participants (PWPD and healthy adults) and communication partners can be found in Table 8. The ICC were only calculated for the PWPD, as preliminary analyses showed no correlation between the judgment of the controls and their communication partners. The judgment of the communication partners had a fair to excellent agreement to that of the PWPD for all subtests. The ratings of the communication partners were most similar to those of the PWPD on the BigCAT (ICC=0.795) and SSC-SD (ICC=0.767). For the SSC-ER the agreement was good (ICC=0.713), but only fair for the BCL (ICC=0.573), indicating more differences in the ratings of PWPD and their communication partners.

## Table 8

Intraclass Correlation between Participant and Communication Partner Judgement for the Subtests of the BAB-Voice

	Intraclass	95% Confidence	Judgment based on
	Correlation	interval	Cichetti (1994)
BCL	0.573	0.275-0.769	Fair
BigCAT	0.795	0.610-0.897	Excellent
SSC-ER	0.713	0.484-0.851	Good
SSC-SD	0.767	0.571-0.881	Excellent

Note. The two-way mixed, absolute agreement, single measures Intraclass Correlations were

reported.

Abbreviations. BCL, Behavior Checklist; BigCAT, Communication Attitude Test for Adults;

SSC-ER, Speech Situation Checklist – Emotional Reaction; SSC-SD, Speech-Situation

Checklist – Speech Disruption
The difference between the scores given by the PWPD themselves and those given by the communication partners was also determined using a mixed MANOVA. There was no effect of rater (PWPD vs. proxy) on the subtest scores of the BAB-Voice (Pillai's trace = 0.247, F[4]=2.216, p=0.094,  $\eta_p^2$ =0.247), which showed that the ratings of PWPD and their communication partners did not differ significantly.

# Research Question 4: Relationship between the affective, cognitive, and behavioral reactions to hypophonia in PWPD

The relationships among the scoring different BAB-Voice subtests themselves were considered using Pearson's correlations. All subtests correlated with one another to a high degree. The highest correlation was found between the SSC-ER and the SSC-SD subtest (r[48]=0.94, p<0.001). This indicated that the emotional reactions and voice disruptions in different speech situations were closely related to each other. The communication attitude, as described by the BigCAT, also was highly related to all other dimensions: emotional reaction (SSC-ER, r[48]=0.82, p<0.001), voice disruptions (SSC-SD, r[48]=0.81, p<0.001), as well as the used coping behaviors (BCL, r[48]=0.78, p<0.001). The smallest correlations were found between the coping behaviors (BCL), the experienced voice difficulty (SSC-SD, r[48]=0.75, p<0.001) and the emotional reaction (SSC-ER, r[48]=0.74, p<0.001).

#### **Research Question 5: Internal Consistency of BAB-Voice in PWPD**

Finally, the internal consistency of all four of the subtests was examined using ICC. Two-way mixed, consistency, average measures ICC were employed and can be found in Table 9. Based on Cichetti's (1994) criteria, the items of the BCL were in excellent agreement with each other, as were those of the BigCAT, SSC-ER and SSC-SD.

#### Table 9

	Intraclass	95% Confidence	Judgment based on
	Correlation	interval	Cichetti (1994)
BCL	0.882	0.830-0.924	Excellent
BigCAT	0.970	0.956-0.980	Excellent
SSC-ER	0.992	0.988-0.995	Excellent
SSC-SD	0.991	0.986-0.994	Excellent

#### Intraclass Correlation for the Items on Subtests of the BAB-Voice

*Note.* The two-way mixed, absolute agreement, average measures Intraclass Correlations were reported.

*Abbreviations*. BCL, Behavior Checklist; BigCAT, Communication Attitude Test for Adults; SSC-ER, Speech Situation Checklist – Emotional Reaction; SSC-SD, Speech-Situation Checklist – Speech Disruption

## Discussion

The dissertation study aimed to describe how PWPD feel, think, and act in reaction to the experienced voice difficulties during the disease process using the BAB-Voice. Specifically, the affective, cognitive, and behavioral reactions to voice use were compared in PWPD and healthy controls. Additionally, the self-judgment of PWPD was compared to that of a daily communication partner. The subtest scores of PWPD on the BAB-Voice were significantly higher than those of the controls, indicating a larger perceived psychosocial impact on communication. In addition, the ratings of daily communication partners of PWPD were in agreement with the ratings of the PWPD themselves.

PWPD scored significantly higher than healthy adults on all the subtests of the BAB-Voice, indicating more experienced voice disruption, negative feelings and attitude, and more use of coping behaviors. These results confirmed that the hypokinetic dysarthria experienced in PWPD could give rise to psychosocial consequences. Existing literature has reported that PWPD may feel frustrated or embarrassed about their speech (Johansson et al., 2020; Whitehead, 2010; Yorkston et al., 2017) or that they may experience difficulty with communication in certain circumstances or with certain people (Johansson et al., 2020; Schalling et al., 2017; Whitehead, 2010; Yorkston et al., 2017). The communication changes could even lead to coping behaviors such as avoiding or changing speech (Schalling et al., 2017; Whitehead, 2010). The results of the current study indicated that the BAB-Voice was able to quantify the situational voice difficulty and negative feelings (SSC-SD, SSC-ER), negative attitude regarding communication (BigCAT), and coping behaviors (BCL) related to the hypophonia in PD.

The findings from this study are aligned with those from previous reports which studied the psychosocial consequences of hypokinetic dysarthria. Letanneux et al. (2013) and Cardoso et al. (2018) considered the Dysarthria Impact Profile in PWPD. Both studies found significantly lower scores on the Dysarthria Impact Profile in PWPD, indicating a larger psychosocial impact of communication difficulties in PWPD. Similarly, for the Communicative Effectiveness Survey, multiple studies described lower scores on the scale in PWPD (Donovan et al., 2008; Dykstra et al., 2015). These lower scores reflected more issues with communicative participation compared to healthy adults (Donovan et al., 2008; Dykstra et al., 2015). While the current study considered additional related measures involving the psychosocial impact of hypokinetic dysarthria, the collective results of this investigation along with existing literature suggest that PWPD experience more psychosocial consequences associated with vocal changes than do people without the disease. Collectively,

the results from these studies support that the hypokinetic dysarthria and hypophonia impact the psychosocial or participatory well-being of PWPD and that these changes are measurable in questionnaire-based instruments.

It is of note that the subscores for the BAB-Voice subtests were highly variable, especially for PWPD. The clinical profile of PD is highly variable (Wolters, 2008), including the extent of hypokinetic dysarthria present from one speaker to the next (Majdinasab et al., 2016; Sapir, 2014). As the presence of a voice disorder was not a prerequisite to participate in the current study, some PWPD could have experienced voice difficulty and consequently did not feel impaired because of their voices, while other PWPD may have experienced substantial vocal changes. Additionally, the sample of healthy adults may not have been completely normophonic. Dysphonia is present in a substantial part of the aging population (Marino & Johns, 2014). Despite the presence of dysphonia in the older population, it is often not diagnosed or treated, as people consider the vocal changes a normal aspect of aging (Marino & Johns, 2014). One example of a typical voice disorder in an aging population is presbyphonia, the vocal changes due to aging. People with presbyphonia would present with a hoarse and breathy voice, lower vocal intensity, vocal fatigue, and vocal instability (Galluzzi & Garavello, 2018), symptoms similar to the hypophonia in PWPD. Therefore, it would be possible that some healthy adults included in our control example did present with some kind of voice disorder. However, it was impossible to verify the presence of unreported dysphonia in the control subjects, or the extent of voice disorders in PWPD, as no voice recordings were acquired.

The results of the current study could be compared to other populations who have been assessed with the BAB-Voice, such as the version of the BAB-Voice administered in the study by Watts and Vanryckeghem (2017). In that study, the BAB-Voice was used to measure the emotional, cognitive, and behavioral reactions in a population with SD (Watts & Vanryckeghem, 2017). Similar to Parkinson's Disease, SD is a neurological voice disorder (Hintze et al., 2017). However, it is a focal neuropathology (e.g., localized at the larynx, Hintze et al., 2017) and not a neurodegenerative disease like PD. The exact pathophysiology of SD remains unclear (Hintze et al., 2017). The participants with SD overall scored higher than PWPD on all subtests of the BAB-Voice. In the SD sample, participants achieved scores close to the maximum scores of the subtests (Watts & Vanryckeghem, 2017). On the other hand, the sample of PWPD scored in the mid-to-low ranges of each subtest. The scores on the BigCAT-subtest could also be compared with other samples of people with SD using a different version of the BAB-Voice (Vanryckeghem et al., 2016; Vanryckeghem & Ruddy, 2015), and with PWS as the BigCAT items in the BAB (Vanryckeghem & Brutten, 2011) were almost identical to those in the BAB-Voice. The samples of people with SD also scored markedly higher than the PWPD (Vanryckeghem et al., 2016; Vanryckeghem & Ruddy, 2015). While the PWS scored similarly to the people with SD, the scores of the PWPD were once again lower on the BigCAT. These results could indicate that PWPD experience less emotional, cognitive, and behavioral reactions to their speech disorder than people with SD, and less cognitive reactions than PWS do. However, both SD and stuttering are disorders that interrupt the normal flow of speech (Vanryckeghem et al., 2016; Vanryckeghem & Ruddy, 2015). While dysfluencies are reported in PWPD, the more prevalent and salient characteristics of hypokinetic dysarthria are of a less transient nature (Benke et al., 2000; Miller, 2017; Ramig et al., 2004). The presence of predominantly sudden, unpredictable interruptions of speech in PWS and people with SD possibly could be perceived as more

debilitating than the more stable voice changes seen in PWPD. Moreover, the current study specifically emphasized the psychosocial impact of voice disorders in PWPD. As discussed above, voice disorders were only a single component of the speech difficulty seen in PWPD. While hypophonia is a salient characteristic of voice in PWPD (Miller, 2017), it might not be the most debilitating aspect of hypokinetic dysarthria. Administering a similar instrument focusing on the hypokinetic dysarthria as a whole could therefore yield different results.

In agreement with previous BAB-Voice-research (Vanryckeghem & Ruddy, 2015), the different subtests of the BAB-Voice correlated with each other to a high degree in the current sample. This indicated that the experienced voice difficulty, negative emotions, negative attitude, and coping behaviors were related to each other. Especially the experienced negative emotion and voice difficulty were highly related with each other, as in Vanryckeghem and Ruddy's (2015) study, likely due to the similarities between both subtests. The presence of negative attitudes was also highly related to the experienced voice difficulty, emotional reaction, and coping behaviors. The final aspect considered on the BAB-Voice in PWPD was its internal consistency. The initial analysis showed promising results, as the ICC values were high for all subtests. This indicated that within each BAB-Voice subtest all items were in good agreement, and thus likely tested the same construct. Both the BAB (Svenning et al., 2021; Węsierska et al., 2018) and the BAB-Voice (Vanryckeghem et al., 2016) have previously shown similar results for their internal reliability.

The current sample showed fair to excellent agreement and no significant difference between the ratings of the communication partner and the PWPD on the BAB-Voice, indicating that the communication partners were able to gauge the psychosocial impact

PWPD experience due to their voice disorders. It is of note that while the communication partners could estimate the communication attitude and speech disruption very well, they did not agree as much for the coping behaviors and experienced emotional reactions, indicating these reactions may be harder to gauge. The BAB was used previously to compare the results of PWS with their life partners (Svenning et al., 2021). Svenning et al. (2021) found that while life partners underestimate the impact of stuttering on the person who stutters, the life partner and person who stutters did rate the emotional impact of the speech difficulty in a comparable way. They found positive correlations between partner ratings for the SSC-ER, SSC-SD, and BigCAT. The ratings of SSC-ER and SSC-SD were significantly different between PWS and their life partners, but for the BigCAT they were not (Svenning et al., 2021). The current sample showed a similar trend: the scores of the PWPD typically showed numerically higher values, though there were no significant differences between how the communication partners scored the BCL, BigCAT, SSC-ER, and SSC-SD. Miller et al. (2008) and Parveen and Goberman (2017) found similar results in PWPD using different tools: caregivers tended to rate communication more positively but their assessment did not differ significantly from that of the PWPD. Likewise, Dykstra et al. (2015) found no differences between the scores of PWPD and their communication partners on the Communicative Effectiveness Survey. On the other hand, Donovan et al. (2008) found that the score of PWPD on the Communicative Effectiveness survey was significantly higher for PWPD compared to their communication partner, indicating that PWPD rated themselves as more disabled. Overall, the literature suggests that communication partners can provide some indication of what the individual with PD is experiencing. Including the assessment of communication partners is important for several reasons. While patient report remains the

most important source of information, communication partners can provide clinically valuable information when the PWPD would be unable to due to physical and/or cognitive limitations (Dykstra et al., 2015; Miller et al., 2008; Parveen & Goberman, 2017). They can also provide insight into the PWPD's communicative functioning outside of the clinical setting (Donovan et al., 2008). Miller et al. (2008) even suggest that having a frequent communication partner or informed listener may be beneficial to the PWPD, as communication partners can ease some of the communicative burdens. Consequently, incorporating communication partners of PWPD during both the diagnostic and treatment process of hypokinetic dysarthria and hypophonia may be beneficial, as communication partners are often involved in communication with the PWPD.

The current study also aimed to compare two different modes of administration for the BAB-Voice. For the sample within the DFW Metroplex, a clinician administered the instrument. Participants from other locations in the United States were invited to fill out the BAB-Voice by themselves. The current study did not find significant differences between the two modes of administration. These results could inform how the tool is administered in the clinic. Given that no significant differences were found between one mode of administration or the other, clinicians could be flexible with the used mode of administration. The clinician could choose the mode of administration that best suits the capability and need of both the patient and clinician. From the experience during this research project, the clinician-guided mode of administration had the advantage that the participant could ask for more clarification while filling out the tool in real-time. On the other hand, given that the BAB-Voice is an extensive instrument, it could be more time-efficient to ask the participants to fill out the instrument in a self-guided manner before the (research) appointment. In the literature, little

information was available on how the mode of administration could potentially influence an instrument's outcome in the literature. The current study was a first step in determining if different administration modes can influence participants' replies on questionnaire-based instruments.

As discussed above, the value of some instruments measuring aspects of the psychosocial impact of the communication changes in PWPD has been established (Baylor et al., 2013; Cardoso et al., 2018; Dykstra et al., 2015; Letanneux et al., 2013). The current study shows that the BAB-Voice has the potential to be a reliable tool to measure the psychosocial impact of vocal changes in PWPD. Moreover, the BAB-Voice gives a more extensive perspective on a person's communicative functioning (Vanryckeghem et al., 2016). It specifies the voice difficulty and anxiety in detailed speech situations. It also quantifies specific coping behaviors and communicative attitude (Vanryckeghem et al., 2016). The BAB-Voice subtests can aid clinicians in determining more multimodal, holistic treatment approaches as well as specific targets within treatment (Vanryckeghem et al., 2016). Given that the psychosocial impact of the hypokinetic dysarthria is typically not treated in PWPD, this information can inform new treatment approaches.

The current study presents several limitations. Firstly, the size of both the clinicianguided sample and the participant-guided sample was small. Moreover the PWPD and control groups were not balanced. One possible explanation for the difficulty recruiting is the presence of COVID-19 during the time of recruitment. Moreover, some of the participants in the considered population (PWPD, and adults older than 50) indicated they did not have a daily communication partner and thus were excluded from the study. The length of the BAB-Voice, and thus the time needed to fill out the instrument, may also be a contributing factor

to the incomplete responses or non-participation of some prospective participants. In future research, allowing PWPD or controls to participate by themselves or splitting the administration of the subtests over different days could be considered to promote larger sample sizes. Moreover, a longer recruitment period and more targeted recruitment of controls could result in a more balanced sample. While the participant-guided sample (collected in DFW-area) and the clinician-guided sample (collected in the USA, but outside DFW) were found to not differ for the considered parameters, both samples could still differ for other variables not presently considered. Moreover, the PWPD and healthy controls did differ significantly for age. While the MANCOVA analysis did control for age when comparing the subtest scores, both samples could still differ from one another for other variables. Another limitation was that the current study only considered vocal symptoms. As described in the introduction, PWPD present with hypokinetic dysarthria, which affects all subsystems of speech (Brabenec et al., 2017; Magee et al., 2019; Ramig et al., 2004). Consequently, participants could have considered other speech-related symptoms not related to hypophonia when filling out the assessment tool. While a general explanation regarding voice at the start of the BAB-Voice, as well as reminders, were provided in the instrument, these measures could not guarantee that participants only considered vocal symptoms. Future research could focus on the hypokinetic dysarthria in PWPD as a whole, or consider ways to ensure participants solely consider voice-related symptoms while filling out the BAB-Voice. Finally, due to the COVID-19 constraints during data collection, collecting speech samples from the participants filling out the instrument was impossible. In the future, a research project may be conducted that compares the voice quality (as measured acoustically and

perceptually) in PWPD with the taken psychosocial measures to see if any correlations exist between the two.

### Conclusion

The current study aimed to describe the affective, behavioral, and cognitive reactions to the vocal changes in Parkinson's Disease using the BAB-Voice. PWPD indicated that they experienced significantly more negative emotions and voice disruption in different speech situations compared to healthy adults. PWPD also used significantly more coping behaviors and showed more negative attitude regarding their voice. Communication partners largely agreed with the judgment of the PWPD, indicating they are aware of the difficulties the PWPD are experiencing.

#### V. Discussion

# Description and Diagnosis of Voice Disorders and their Psychosocial Consequences General Voice Disorders

**Voice Disorder Assessment in Practice.** Voice disorders lead to deviating voice quality (Aronson & Bless, 2011; Ramig & Verdolini, 1998), which is a multidimensional perceptual description of voice (Barsties & De Bodt, 2015). SLPs use multiple measures to describe and diagnose voice quality and disorders (Dejonckere, 2010). This analysis aims to inform the therapist's treatment plan (Behrman, 2005; LeBorgne & Donahue, 2019) and to determine the effect of treatment (Dejonckere, 2010). The typical voice assessment protocol describing voice quality contains five distinct elements: auditory-perceptual evaluation of the voice signal, acoustic analysis of a voice recording, aerodynamic measures, visualization of the vocal folds, and self-perceived voice quality (Barsties & De Bodt, 2015; Dejonckere, 2010; LeBorgne & Donahue, 2019).

Auditory-perceptual assessment of voice is often considered the "gold standard" for describing voice and voice quality and is instrumental in determining the presence of a voice disorder (Carding et al., 2009; Kempster et al., 2009; Oates, 2009). Perceptual assessments require an expert listener to indicate how a voice differs from normal for multiple parameters (Carding et al., 2009). Given that voice is a perceptual phenomenon, auditory perceptual evaluation is an intuitive choice to evaluate voice (Oates, 2009) that is also cost- and timeefficient (Barsties & De Bodt, 2015; Carding et al., 2009; Oates, 2009). Consequently, SLPs commonly use perceptual evaluation in voice practice (Oates, 2009). Over the years, researchers have developed several scales to improve the standardization and reliability of perceptual voice measures (Dejonckere, 2010). One example of a commonly used tool to

perceptually rate voice quality is the Consensus Auditory-Perceptual Evaluation of Voice (CAPE-V, Kempster et al., 2009). The CAPE-V protocol is a standardized tool and consists of a fixed set of stimuli (sustained vowel – sentences – conversation) that are rated using a visual analog scale for six different features: overall severity, roughness, breathiness, strain, pitch, and loudness (Kempster et al., 2009). Clinicians then convert the visual analog scale to a number ranging between 0 – 100. Other scales, such as the GRBAS-scale (Hirano, 1981) are available in both practice and research as well.

While auditory-perceptual measures have been criticized for being subjective and having issues with reliability, acoustic voice measures are typically considered to be more objective (Barsties & De Bodt, 2015; Dejonckere, 2010; Ziethe et al., 2011). While singular acoustic analyses (e.g., perturbation analysis, cepstral based measures, etc.) can be performed on sustained vowels or running speech (Dejonckere, 2010; Maryn, De Bodt, et al., 2010), better reliability and validity has been found when multiple acoustic parameters are combined in one index (Barsties & De Bodt, 2015). The Acoustic Voice Quality Index (Maryn, Corthals, et al., 2010; Maryn, De Bodt, et al., 2010) is an example of such a multiparametric approach. The AVQI is calculated using multiple acoustic measures based on spectral, cepstral, and time-based measures taken from both sustained vowels and continuous speech (Maryn, Corthals, et al., 2010; Maryn, De Bodt, et al., 2010). The measure then results in a score ranging from 1 to 10, with a higher number indicating greater dysphonia (Maryn, Corthals, et al., 2010; Watts & Awan, 2019). Regardless of the measures used in the analysis, the reliability of the acoustic voice measures relies heavily on the equipment and programs used, as well as the environment in which the voice samples are collected (Barsties & De Bodt, 2015).

Aerodynamic measures focus on the respiratory driving force behind phonation (Ziethe et al., 2011). Clinicians typically measure airflow, air pressure, and/or lung volumes for voice diagnostics (Barsties & De Bodt, 2015; Ziethe et al., 2011). Some aerodynamic measures, such as the maximum phonation time, are widely used and require little to no effort or equipment (Dejonckere, 2010). Other aerodynamic measures, such as vital capacity, average flow rate during phonation, or phonation threshold pressure, require more specialized equipment as spirometers or pneumotachographs (Dejonckere, 2010; Ziethe et al., 2011). These aerodynamic measures correlate closely to vocal fold physiology and thus provide important information related to vocal fold pathology (Ziethe et al., 2011).

The visualization of the vocal folds is considered an essential element in diagnosing voice disorders, as neither auditory-perceptual, acoustic, or aerodynamic measures can provide disorder-specific clues for diagnosis (Dejonckere, 2010; Ziethe et al., 2011). Videostroboscopy is the standard to analyze vocal fold vibration (Ziethe et al., 2011). This technique uses a visual illusion to "slow down" the vocal folds, and thus allows for the subjective rating of vocal fold vibration (Ziethe et al., 2011). Parameters measured during these ratings are the symmetry, amplitude, and periodicity of the vocal vibration, as well as the mucosal wave and the closure pattern (Dejonckere, 2010; Ziethe et al., 2011).

The last part of voice disorder assessments, self-perceived voice quality, takes into account the person's own opinion on their voice and the experienced voice difficulty. It aids in describing the voice difficulty, disability, and handicap a person experiences during different aspects of their daily life, rather than being a snapshot of what happens in the voice clinic (Carding et al., 2009; Dejonckere, 2010; Ziethe et al., 2011). Patients can assess their voices using multiple instruments that have been developed over the years (Carding et al.,

2009; Dejonckere, 2010; Ziethe et al., 2011). A commonly used and researched tool is the Voice Handicap Index (VHI), which measures the psychosocial handicap that occurs due to voice disorders (Jacobson et al., 1997). The VHI consists of 30 questions, spread out evenly over three subscales: functional, physical, and emotional (Jacobson et al., 1997). The patient answers each question on a scale from 0 (never) to 4 (always) (Jacobson et al., 1997). The advantage of the VHI is that it has been translated into multiple languages and that it has been adapted to specific populations with voice disorders (Ziethe et al., 2011).

Voice Disorders within the International Classification of Function, Disability, and Health. All of the above-mentioned measures function within a larger framework. The World Health Organization (WHO) developed the International Classification of Functioning, Disability, and Health (ICF) to provide a standardized concept and framework to describe and measure disability (World Health Organization, 2002, 2013). ICF put health and functioning into a multi-dimensional framework (World Health Organization, 2013). According to the ICF, a person's functioning and disability are the consequence of how the health condition interacts with the person and the environment. The ICF is, therefore, split in two: one part covers "Functioning and Disability", the other "Contextual Factors." The "Functioning and Disability" factor consists of "Body Function" (i.e., body physiology), "Body Structures" (i.e., body anatomy), and "Activities and Participation" (i.e., execution of tasks and involvement in life situations), whereas "Contextual Factors" cover "Environmental Factors" (i.e., the physical, social, and attitude-related circumstances) and "Personal Factors" (not specifically coded within ICF) (World Health Organization, 2013). Describing patients using these elements can lead to more holistic assessment and treatment of patients (World Health Organization, 2013).

The American Speech-Language-Hearing Association has adopted the ICFframework within the scope of speech-language pathology to more comprehensively describe patient's communication disorders and functionality (American Speech-Language-Hearing Association, n.d.). Multiple authors have described how the ICF can be used within communication sciences and disorders (Cruice, 2008; Mccormack & Worrall, 2008; Threats, 2006; Threats & Worrall, 2004). Specifically for voice disorders, Ma et al. (2007) have described how clinicians and researchers can employ the ICF. The ICF-framework has the advantage that it centers communication within broader life skills (Threats & Worrall, 2004). It consequently has the potential to guide both clinical and research-based endeavors while focusing on the individual's needs (Cruice, 2008; Threats, 2006).

The "Body Structures" considered in voice disorders are the anatomical changes, primarily within the larynx or the cranial nerves, and are typically assessed with the abovementioned techniques to visualize the larynx (E. P. M. Ma et al., 2007). On the other hand, the "Body Functions" cover both the voice production and voice quality, as well as the emotional and personality-related function of a person. To measure the body functions related to voice, SLPs can use auditory-perceptual measures (E. P. M. Ma et al., 2007), as well as acoustic and aerodynamic measures. "Activities and Participation" specifically describe the person's functional status. This category gives more information on a person's limitations in different speech situations such as conversation, phone calls, social events, or even job functions (E. P. M. Ma et al., 2007; Threats, 2006; Threats & Worrall, 2004). Most of the patient's self-assessment tools, such as the VHI, consider these areas (E. P. M. Ma et al., 2007). While these tools give an idea of the experienced difficulties with activity and participation, they may not provide a comprehensive description. The "Environmental

Factors" consider what support (e.g., voice amplifiers, ability to take time off) or barriers (e.g., noise, negative attitudes toward voice disorders) are apparent in the environment in which the voice-disordered person has to function (E. P. M. Ma et al., 2007). Moreover, these environmental factors are typically outside the person's own control (Threats, 2006). Other areas that might influence the functioning of a person, the "Personal Factors," are aspects as gender, age, and race, as well as lived experiences, personality, coping style, and lifestyle (Cruice, 2008; E. P. M. Ma et al., 2007). The individuality of the considered person, along with their wants and needs, have the potential to influence how a person reacts to the health condition and its treatment (Cruice, 2008; Mccormack & Worrall, 2008). SLPs can address the "Contextual Factors" of the ICF-model during anamnesis while making up the casehistory or through observation, but typically do not separately test them (E. P. M. Ma et al., 2007). Historically, SLPs have focused primarily on the "Body Functions" and "Body Structures" (Mccormack & Worrall, 2008). While some tools for the "Activities and Participation" area exist, these are general (e.g. VHI) and do not provide enough information to inform specific treatment goals.

#### Voice Disorders in Parkinson's Disease

Speech and voice disorders are apparent in PWPD throughout the disease process (A. Ma et al., 2020; Ramig et al., 2004; Skodda et al., 2013). These changes are thought to be the consequences of central processing issues, and possibly the dopamine deficiency during the later stages, brought about by the PD disease (A. Ma et al., 2020; Miller, 2017; Sapir, 2014). Clinicians and researchers typically assess hypophonia, the vocal symptoms in PD, using the above-described voice quality outcome measures. However, as mentioned in the introduction, the majority of the information on hypophonia in PD is based on PWPD in the

advanced stages of the disease. Moreover, the findings on the early or non-advanced stages of PD are difficult to compare as researchers use different definitions for these terms. Nevertheless, the following section discusses the available information on hypophonia in different stages of the disease, focusing on the clinically useful measures. The description and assessment of the hypophonia throughout the PD process are important to determine the optimal treatment goals and timing. Especially the description of early-stage hypophonia is important, as early-stage interventions could potentially incur greater gains in voice quality (Boutsen et al., 2018; Gibbins et al., 2017).

Auditory-Perceptual Characteristics of Hypophonia. The literature has described the auditory-perceptual characteristics of voice in advanced-stage PWPD in detail. Hypophonia is a collection of multiple perceptual characteristics. PWPD's voices typically show reduced loudness, as well as a breathy, rough, and hoarse voice quality (Holmes et al., 2000; A. Ma et al., 2020; Ramig et al., 2004; Stewart et al., 1995). The voice may lose its variability resulting in monopitch and monoloudness (Holmes et al., 2000; A. Ma et al., 2020; Ramig et al., 2004). Moreover, vocal tremor may be audible in the voice signal (Ramig et al., 2004), but is possibly only present in the late stages of PD (Holmes et al., 2000). For example, Midi et al. (2008) compared the voices of PWPD with those of healthy controls using the GRBAS scale and found that PWPD presented with significantly more breathiness and asthenia in their voices. Similarly, Bauer et al. (2011) and Castro et al. (2020) found increased ratings of all aspects of the GRBAS scale in PWPD. One limitation of the available literature is that some of the authors employ their own perceptual scales (e.g., Cushnie-Sparrow et al., 2018; Holmes et al., 2000) rather than the standardized scales typically used in voice clinics.

While vocal dysfunction has been cited as one of the early signs of Parkinson's Disease (A. Ma et al., 2020; Ramig et al., 2004), it is unclear if clinicians can perceive these early vocal changes. Lirani-Silva et al. (2015) did not find perceptual differences for phonation when comparing PWPD with adults but used a very general instrument to assess voices. On the other hand, compared to controls, PWPD in early stages have been found to present with more breathiness, harshness, monopitch, and monoloudness, though to a lesser extent than the late-stage PWPD (Holmes et al., 2000; Stewart et al., 1995). Lechien et al. (2020) found significantly worse ratings of all GRBAS-factors but roughness in early-stage PD. These perceptual changes are similar to the ones described in Chapter III, which aimed to contribute to and clarify the description of hypophonia in non-advanced disease stages.

Chapter III described the perceptual characteristics of Parkinsonian voices in the nonadvanced stages of the disease, focusing on describing the "Body Functions" of PWPD. This study, therefore, focused on the most typical way of assessing voice, using auditoryperceptual evaluation. Two clinicians specialized in voice perceived the voices of PWPD as more severely dysphonic, more breathy, and older than those of healthy controls. However, both group's voices were perceived as equally rough. Given that the project employed an adapted version of the CAPE-V scale, the found results can easily be translated to use in a voice clinic. While this study contributes to the body of literature on voice in early PD, many questions remain. Overall, the literature indicates that vocal changes are perceivable, even in the non-advanced stages of PD. However, the predissertation study as well as the existing literature do not indicate when the acoustic changes are audible or when PWPD may start noticing them. Moreover, it is unclear if these initial changes are also apparent in the vocal

fold anatomy and physiology, or the aerodynamic measures. The below section will discuss what is known on both late-stage and early-stage hypophonia for those aspects.

Acoustic Characteristics of Hypophonia. The voice signal with salient hypophonia will contain more noise and be less regular, resulting in increased jitter and shimmer, and reduced harmonics-to-noise ratios (Castro et al., 2020; Jiménez-Jiménez et al., 1997; A. Ma et al., 2020; Midi et al., 2008; Ramig et al., 2004; Skodda et al., 2013; Yücetürk et al., 2002). The intensity (or sound-pressure level) becomes lower, and both intensity variability and frequency variability become smaller (A. Ma et al., 2020; Oguz et al., 2006; Ramig et al., 2004; Yücetürk et al., 2002). Higher fundamental frequencies have also been detected acoustically (A. Ma et al., 2020) along with vocal tremor (Gillivan-Murphy et al., 2019a). In the later stages of the disease or patients with considerable hypophonia, the clinically-used acoustic voice measures may lose their ability to describe the voice quality of PWPD, as PWPD's voices become too aperiodic to achieve reliable measures (Brabenec et al., 2017).

Specific to the early stages of PD, researchers have used acoustic voice analysis as a possible prodromal marker of PD (Brabenec et al., 2017). Brabenec et al. (2017) reviewed how acoustic voice and speech parameters could help diagnose PWPD in the early stages of PD. Factors related to the fundamental frequency, its variability, and prosody were the vocal characteristics that aided most in early diagnosis (Brabenec et al., 2017). When considering more clinically relevant measures, increased values for jitter, shimmer, and noise-to-harmonics ratio in early-stage PD were found (Rusz, Cmejla, Ruzickova, & Ruzicka, 2011; Rusz, Cmejla, Ruzickova, Klempir, et al., 2011). Lechien et al. (2020) found differences for these same parameters, along with increased fundamental frequency, and increased prevalence of vocal tremor in PWPD. These measures indicate an increase in noise and

irregularity in the voice and are expected to deviate in voices with increased perceived breathiness and harshness, which is how the voice in PWPD has been described (see above). However, while these acoustic differences may already be present in the early stages of PD, PWPD do not necessarily perceive them (Fernández-García et al., 2020).

#### Endoscopic and Videostroboscopic Characteristics of Hypophonia. The

videostroboscopic image of Parkinsonian vocal folds shows several characteristics. Generally, more PWPD present with laryngeal abnormalities (either anatomical or physiological) than healthy controls (Yücetürk et al., 2002). Incomplete glottal closure or bowing can be important, as they result in air leakage, which can cause the hallmark perceptual characteristics of Parkinsonian voice (Bauer et al., 2011; Blumin et al., 2004; Castro et al., 2020; A. Ma et al., 2020; Midi et al., 2008; Ramig et al., 2004; Yücetürk et al., 2002). However, not all studies found abnormal closure patterns (Jiménez-Jiménez et al., 1997). Other findings are asymmetrical vibratory patterns (A. Ma et al., 2020; Ramig et al., 2004), reduced or irregular mucosal wave (Bauer et al., 2011; Yücetürk et al., 2002), and arytenoid edema (Castro et al., 2020). In some cases, researchers could observe a laryngeal tremor as well (Bauer et al., 2011; Blumin et al., 2004; Castro et al., 2020; Jiménez-Jiménez et al., 1997; Midi et al., 2008; Ramig et al., 2004).

On the other hand, the visualization of the vocal folds in the early stages is not well described in the literature. Gibbins et al. (2017) suggested that in the early stages, no laryngeal changes were apparent. By the mid-stage PWPD developed compensatory muscle changes, which then disappeared in the final stage, when the laryngeal abnormalities became pronounced. More research is needed on this topic.

Aerodynamic Characteristics of Hypophonia. PD also affects respiratory function (Torsney & Forsyth, 2017), which is important for voice production. Ramig et al. (2004) found lower vital capacities in PWPD. The maximum phonation time of PWPD is shorter than that of healthy controls (Bauer et al., 2011; Midi et al., 2008; Ramig et al., 2004; Yücetürk et al., 2002). Moreover, abnormal airflow patterns can be present in PWPD (Ramig et al., 2004). For example, less phonatory resistance (Hammer & Barlow, 2010; Motta et al., 2018), larger airflow during vowel production (Burk & Watts, 2018), longer open quotients (Matheron et al., 2017), and decreased subglottal pressure (Hammer & Barlow, 2010; Matheron et al., 2017) have been described. Burk and Watts (2018) suggested that the deviance of airflow measures could depend on the motor phenotype in PD, with the nontremor dominant type experiencing more difficulties.

Not much data is available on the respiratory measures in early-stage PD, but overall the literature seems to indicate that aerodynamic changes could be a later symptom of vocal dysfunction in PD. Motta et al. (2018) considered PWPD with little dysphonia and did not find significant differences for many of their considered aerodynamic parameters. Similarly, Lechien et al. (2020) did not find significant differences for maximum phonation time, vital capacity, and the phonatory quotient in early-stage PD.

Self-Assessment of the Psychosocial Consequences of Hypophonia. The abovediscussed assessments of voice focus on the "Body Structures" and "Body Functions" within the ICF-framework. The last aspect of a typical voice assessment covers "Activities and Participation". The VHI (Jacobson et al., 1997) is a standard tool used in voice evaluations and is a feasible, reliable, and valid tool in PWPD (Guimaraes et al., 2017). The VHI was able to distinguish between PWPD with voice difficulty and those without the disease

(Guimaraes et al., 2017). Moreover, researchers have found elevated scores on the VHI for PWPD compared to healthy adults (Bauer et al., 2011; Castro et al., 2020; Gillivan-Murphy et al., 2019a; Midi et al., 2008; Motta et al., 2018). As mentioned above, the VHI describes the psychosocial consequences spread out over functional, physical, and emotional subscales (Jacobson et al., 1997). While the advantage of the VHI is that it is a generally accepted and widely used tool, it does not provide detailed information on the psychosocial consequences in PWPD. For example, it does not give information on the effect of different speech situations on the vocal function, emotion, and participation, or on the use of coping behaviors. Nevertheless, PWPD have indicated that they do experience these psychosocial difficulties in their daily lives as a consequence of the hypokinetic dysarthria and hypophonia. They often experience negative feelings of frustration, anxiety, and fatigue (Johansson et al., 2018; Whitehead, 2010; Yorkston et al., 2017), resulting in speech difficulty along with loss of participation in specific situations (Schalling et al., 2017; Whitehead, 2010; Yorkston et al., 2017). PWPD have also reported coping behaviors (Johansson et al., 2020; Schalling et al., 2017; Whitehead, 2010). Interestingly, the abovedescribed psychosocial impact is independent of factors as disease severity and the experienced voice or speech difficulty (Gillivan-Murphy et al., 2019b; Miller et al., 2008, 2011). Consequently, PWPD with incipient hypophonia may already report severe psychosocial difficulty and vice versa. The reported psychosocial issues, that fall within "Activities and Participation", "Personal Factors", and sometimes "Environmental Factors," are not typically well-described during voice examinations.

Researchers have developed or adapted tools to PWPD to remediate this shortage of information, such as the communicative Effectiveness Survey (Donovan et al., 2008; Hustad,

1999), Communication Participation Item Bank (Baylor et al., 2009; Yorkston et al., 2008), and the Dysarthria Impact Profile (Walshe et al., 2009). However, these tools specifically focus on participation or do not provide detailed situational information. Consequently, as described in Chapter IV, the BAB-Voice, a tool that describes the attitude, feelings, and coping behaviors related to voice in detail, was adapted to a population of PWPD.

The dissertation study, similar to the predissertation study in Chapter III, aimed to describe voice disorders in PWPD. However, this study focused on the self-perception of the patient, rather than perceptual measures. The study specifically aimed to compare affective, behavioral, and cognitive reactions to voice difficulty in PWPD and healthy older controls. The scores on the BAB-Voice were significantly higher for PWPD compared to healthy adults, indicating that PWPD experienced more psychosocial impact of their voice disorders than healthy controls, which was expected given the discussed literature. The advantage of the BAB-Voice over the other tools is that its detailed nature allows to directly inform treatment approaches (discussed below). The usability of the BAB-Voice in PWPD also shows the value of working with tools typically used in other specializations of speech-language pathology. While specializing in one area within communication sciences and disorders is valuable, collaborating with other areas may also be beneficial.

Value of Involving Communication Partners. One aspect that was not discussed yet is the potential value of the assessment of communication partners within the diagnosis of voice disorders and more specifically hypophonia. Given that communication partners directly participate in the communication process, they can provide valuable insights for diagnosis and therapy (Gillivan-Murphy et al., 2019b), both on the voice difficulty PWPD experience in different speech situations, as well as their reactions to that voice difficulty.

Most of the studies included partner assessment on the psychosocial impact of hypokinetic dysarthria, and while these studies found a trend for communication partners to underestimate the experienced difficulty of PWPD, the majority did not find significant differences between partner- and self-assessment (Dykstra et al., 2015; Miller et al., 2008; Parveen & Goberman, 2017). A secondary goal of the dissertation study was to determine the agreement between communication partners and PWPD on the BAB-Voice (see Chapter IV). Similar to the discussed literature, the study found strong agreement and no significant differences between the judgments of communication partners and PWPD. While the person's lived experience remains the most important aspect during the voice assessment, communication partner judgment can prove valuable in cases in which the PWPD is not able to provide accurate information (Dykstra et al., 2015; Miller et al., 2008; Parveen & Goberman, 2017). Moreover, given that PWPD are known to have difficulties with selfperception (Parveen & Goberman, 2017; Sapir, 2014), communication partners may provide essential additional information during voice assessment, e.g. patient's functioning in different speech situations. Especially in pathologies that can affect self-perception, such as in PD, SLPs should consider incorporating communication partners during voice assessment.

#### Summary and Suggestions for Future Research

The assessment of voice disorders in PD follows the pattern of typical voice disorder assessment. However, two areas are underrepresented: the description of voice disorders in the early to non-advanced stages of PD, and the psychosocial impact of voice disorders during any stage of the disease. Nevertheless, detailed description of the voice disorder during all stages is instrumental for treatment. In late-stage PD, the auditory-perceptual, acoustic, videostroboscopic, and aerodynamic measures are well researched. This information is lacking for early-stage PD. Generally, the literature characterizes hypophonia in early-stage PD by breathy, harsh voice quality, as evidenced by increases in the acoustic values for jitter, shimmer, and noise-toharmonics ratio. However, the underlying vocal fold pathology and aerodynamic dysfunction remain unclear. Therefore, future research should inform SLPs about the different clinically relevant characteristics of PD, as well as how they relate to one another, to further the understanding of the vocal changes and their underlying disease processes.

Researchers have not yet extensively considered the psychosocial aspects of voice in PWPD in any of the disease stages. The tools that have been used to assess the psychosocial impact all show that PWPD experience a greater impact of the experienced voice difficulty than healthy older controls do. The factors that are most influential in determining the impact of psychosocial factors in PWPD are unclear. More research is needed to further quantify and describe these reactions throughout the disease process to determine if and how clinicians can include these aspects during voice therapy.

Concluding, the description of hypophonia in PWPD is all too often unidimensional in research. While both the typical assessment protocol of voice, along with the ICFframework, require SLPs to provide multidimensional or even holistic measures, current research does not reflect this approach. More studies that address all areas impacted by voice disorders are needed to allow for the comparison and association of the different assessment types.

#### **Treatment of Voice Disorders**

#### **Treatment of General Voice Disorders**

The above-described voice assessment is essential to determine the treatment approach for persons with voice disorders (Dejonckere, 2010; LeBorgne & Donahue, 2019). Behavioral voice treatment is an important aspect of treating voice disorders (Desjardins et al., 2017; Ramig & Verdolini, 1998). Voice therapy can generally be divided into direct and indirect voice therapy, which SLPs can use separately or together (LeBorgne & Donahue, 2019). Direct voice therapy refers to therapeutic exercises directly targeting vocal anatomy, vocal behavior, and their underlying components (LeBorgne & Donahue, 2019; Van Stan et al., 2015). It can consist of a multitude of techniques, for example, vocal function exercises, resonant voice therapy, flow phonation, etc. (LeBorgne & Donahue, 2019). Gartner-Smidt et al. (2013) found that this component takes up to 80% of a voice therapy session. The effectiveness of different direct treatment techniques is typically investigated using randomized controlled trials. A systematic review by Desjardins et al. (2017) found that voice therapy techniques lead to improvements in the above-described assessments. Improvements in self-assessment and acoustic analysis were most commonly found (Desjardins et al., 2017). Longitudinal follow-up showed that the patient's voice quality remained improved post-voice therapy compared to baseline up to at least one month.

Indirect voice therapy, on the other hand, does not immediately tackle the disordered voice behavior, but consists of counseling and education to promote knowledge and behaviors that benefit voice rehabilitation (Gartner-Schmidt et al., 2013; LeBorgne & Donahue, 2019; Van Stan et al., 2015). This approach can include education on voice production, diet modification, environment modification, relaxation, or even psychosocial

counsel (Gartner-Schmidt et al., 2013), all aspects that correspond with the "Activity and Participation", "Environmental Factors", or "Personal Factors" within the ICF-framework. By itself, indirect voice therapy was found to be less effective than direct voice therapy, and SLPs used it less during voice therapy (Gartner-Schmidt et al., 2013, 2017; Speyer, 2008).

In a typical voice therapy setting, SLPs often use an eclectic voice therapy approach, in which they combine multiple therapeutic approaches (LeBorgne & Donahue, 2019). While the effectiveness of singular therapy techniques is well-researched, research has not addressed the more realistic, ecologically valid approach, which was the aim of another predissertation study (see Chapter II). This study focused on determining what voice pathologies are treated in a private practice specializing in voice, as well as what the outcomes of therapy in this clinic were. Multifactorial diagnoses were the most common within this practice. While a large amount of the assessed patients did not receive treatment, those who did had improved acoustic and self-perceived voice quality post-therapy. No differences between the different diagnoses were seen in the outcome measures. Interestingly, while improvements were found, vocal function did not necessarily return to normal. This phenomenon can be explained by the set goals and discharge criteria and can encourage SLPs to include more factors into therapy. When vocal function cannot return to normal post-therapy, a shift to more indirect voice therapy and optimization of the patient's functioning can be proposed. For example, clinicians can address a negative attitude and the use of coping behaviors, educate family members, and use vocal amplifiers. These factors need to be researched further.

#### Treatment of Voice Disorders in Parkinson's Disease

The treatment of voice disorders in PD is a well-researched area within speechlanguage pathology. The typical treatment of hypophonia in PD employs techniques to overcome the neurological difficulties in the disease. The most known program is Lee Silverman Voice Treatment (LSVT-LOUD). LSVT-LOUD addresses the hypokinesia and central processing issues in PD using principles of motor learning by employing a highintensity program that focuses on increasing vocal intensity and retraining sensory feedback (Sapir et al., 2011). More recently, SPEAK OUT! was developed, a program that encourages PWPD to speak with intent to activate their intact pyramidal pathways (Levitt et al., 2015). The bulk of the effectiveness studies of voice therapy in PD considered LSVT-LOUD. A recent systematic review by Yuan et al. (2020) found that post-LSVT-LOUD, PWPD showed significant improvement in vocal intensity, the typical outcome measure for LSVT-LOUD, as well as in their VHI-scores. Moreover, they found indications that these effects were longterm (Yuan et al., 2020). Yuan et al. (2020) did not provide information on other areas of vocal functioning. However, in the literature, post-therapy improvements in perceived hoarseness and breathiness (Baumgartner et al., 2001) have been described, as well as decreases in the acoustic measures jitter and shimmer (Bryans et al., 2021) and increases in fundamental frequency (Ramig et al., 1995). Videostroboscopically, PWPD improved on glottal incompetence and hyperfunction (Smith et al., 1995). Increases in maximum phonation time and other aerodynamic measures were also described post-LSVT (Ramig et al., 1995; Ramig & Dromey, 1996).

How voice treatment impacts the psychosocial aspects of communication in PWPD is less clear, as the therapy mainly focuses on the body structures and functions. Like Yuan et

al.'s (2020) study indicated, PWPD improved on some measures of psychosocial aspects (i.e., VHI scores). However, this improvement was not a ubiquitous finding as some studies did not find significant differences pre/post-LSVT for VHI (Sackley et al., 2018; Spielman et al., 2007), and other studies found that the improvement of the scores was not maintained during follow-up (Spielman et al., 2011; Wight & Miller, 2015). Ramig et al. (2018) used a tool specifically looking at communicative effectiveness and found significant improvements in communicative effectiveness that were maintained at follow-up. In Chapter IV and earlier in the discussion, it was established that hypophonia significantly affects PWPD's participation, emotions, thoughts, and behaviors. Focusing more on indirect voice therapy, and specifically on the treatment of these psychosocial symptoms, may therefore be beneficial when treating PWPD. At present, no evidence is available on this matter.

Similar to what was found in Chapter IV for general voice disorders, a discrepancy exists between PWPD who experience voice disorders and the PWPD who partake in voice treatment. Schalling et al. (2017) found that while 90% of the PWPD reported at least one speech or communication symptom (with vocal symptoms being the most common), only 45% of the PWPD had received an SLP assessment at some point, and 42% received treatment. Common reasons not to seek speech-language pathology services were that they were not needed, that other problems were prioritized, that the patient did not know about the availability of services, or that healthcare providers had not considered the speech problems. Atkinson-Clement et al. (2019) similarly suggested the explanation that neurologists only refer to SLPs once intelligibility is severely impacted. This lack of referral could lead to PWPD starting therapy later in the disease process, while it has been suggested that therapy during the early stages of PD could yield better results (Boutsen et al., 2018; Gibbins et al.,

2017). Knowing more about the ideal timing of voice therapy during the disease process, as well as close communication with other disciplines could potentially improve these numbers.

#### Summary and Suggestions for Future Research

SLPs commonly use behavioral voice therapy to treat voice disorders. It consists of both direct and indirect treatment approaches. While many randomized controlled trials discern the effect of singular treatment techniques, it is known that SLPs typically employ multiple therapy techniques at once. It is unclear if the used voice therapy techniques are still beneficial when they are not employed by themselves in a highly controlled environment. Therefore, more research is needed on the efficacy of these techniques in realistic therapy settings, including for LSVT in PWPD.

Specifically for PWPD, LSVT is the most common treatment approach. Research consistently describes improvements in intensity post-therapy but the impact of treatment on psychosocial characteristics is less clear. LSVT is a treatment program that focuses predominantly on vocal function. Future research projects could include more therapy aspects that also treat the psychosocial characteristics of the hypophonia (e.g., educating communication partners, making the environment more accessible, including counseling, etc.). The additional benefit of these measures compared to LSVT alone is to be considered. Moreover, the potential benefit of prophylactic or early treatment in PWPD is an interesting research topic, as the literature indicates the potential benefits of early intervention. Aspects as patient motivation and long-term effectiveness are two areas to be considered during these types of treatment.

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## Appendices

## **Appendix 1: Demographic Questions for PWPD**

Gender:

- o Male
- o Female

Age: \_\_\_\_\_

What is your country of origin?

What is your native language?

City and state of Residence:

Highest degree attained:

Name or describe your current (or previous) profession:

Have you been diagnosed with Parkinson's Disease?

- o Yes
- o No

If yes, when were you diagnosed with Parkinson's Disease (month+year)?

Are you currently being treated for Parkinson's Disease?

- o Yes
- o No

If yes, how are you being treated for Parkinson's Disease?

- Medication (please specify in the question below)
- Deep brain stimulation
- Other (please specify in the question below)

If you chose 'medication' or 'other' in the previous question, please specify your treatment in the box underneath. \_\_\_\_\_

Are you currently being treated by a speech-language pathologist for speech or swallowing?

- o Yes
- o No

Have you been treated by a speech-language pathologist for speech or swallowing in the past?

- o Yes
- o No

Are you a member of a Parkinson's Disease support group?

- o Yes
- o No

How much do you exercise during the week?

- $\circ 0 1$  hour per week
- $\circ$  2 3 hours per week
- $\circ$  4 5 hours per week
- More than 5 hours per week

Have you had any other neurological issues (e.g. stroke)?

o Yes

o No

If yes, please specify the neurological disorder in the box below.

Do you require hearing aids?

- o Yes
- o No

Do you have any other speech/language problems other than those related to Parkinson's Disease?

- o Yes
- o No

If yes, please list all current or past speech/language problems:

Have you ever been in therapy for any non-Parkinson's Disease related speech or language problems?

- o Yes
- o No

If yes, please explain what you were seeking therapy for and for how long that therapy lasted.

## **Appendix 2: Demographic Questions for Communication Partners**

Gender:

- o Male
- o Female

Age: \_\_\_\_\_

What is your country of origin?

What is your native language? \_\_\_\_\_

State of residence: \_\_\_\_\_

Highest degree attained: \_\_\_\_\_

Name or described your current (or previous) profession:

Has your communication partner been diagnosed with Parkinson's Disease?

- o Yes
- o No

How long have you known your communication partner?

How often do you talk with your communication partner?

- Less than once a week to once a week
- Two to three times a week
- Four to six times a week
- Seven times a week or more

Do you require hearing aids?

- o Yes
- o No

#### Vita

## ZOË THIJS

Doctoral Candidate <u>zoe.thijs@tcu.edu</u> Davies School of Communication Sciences and Disorders Harris College of Nursing & Health Sciences Texas Christian University

## EDUCATION AND EMPLOYMENT

#### **EDUCATION**

2021 2018	Ph.D. in Health Sciences	Texas Christian University (USA)	
2010	Dissertation title: "Self-Perceived Affective, Behavioral, and Cognitive Reactions Associated with Voice and Communication in People with Parkinson's Disease." Advisor: Dr. Christopher R. Watts		
2017 2016	<b>Master in Speech Language and Hearing Sciences</b> <b>Main Subject Logopaedics</b> <i>Graduated with great distinction</i>	Ghent University (Belgium)	
	Master's thesis title: "Pilootstudie: de validiteit en betrouwbaarheid van het Nederlandstalige CAPS-A protocol bij de perceptuele beoordeling van spraak van schisispatiënten." Advisors: Prof. Dr. Kristiane Van Lierde & Dr. Laura Bruneel		
2016 2013	Bachelor in Speech Language and Hearing SciencesMain Subject LogopaedicsGraduated with greatest distinctionHonours thesis title: "Functional magnetic resonance imagduring phonation in healthy female singers with supraglottAdvisor: Prof. Dr. Sofie ClaeysIn collaboration with: Defrancq C., De Moor M.	s Ghent University (Belgium) imaging pilot study: brain activity glottic laryngeal compression."	
EMPL	OYMENT		

- 2018-present Research Assistant in Laryngeal Function Lab (Dr. Christopher Watts) Texas Christian University Fort Worth, Texas, USA
- 2017-2018 Speech-language pathologist within practice Greet Tavernier Hofstade, Belgium

## SUPPLEMENTARY TRAINING & CLINICAL COMPETENCIES

- 2020 **Hybrid Design and Delivery Training** (Texas Christian University, Fort Worth, USA) Training for online teaching
- 2019 **Phonanium Clinical Assessment of Acoustic Voice Signals** (Fort Worth, USA) Training to acoustically evaluate voice signals with Praat

2018 Training AmpCare ESP (Fort Worth, USA)

Training to use Neuromuscular Electrical Stimulation for swallowing therapy

2017 **Training stuttering therapist – Caroline Moerenhout & Gert Reunes** (Ghent, Belgium) Training in diagnosis and treatment of people who stutter

Trained in flexible laryngeal endoscopy, and rigid laryngeal endoscopy, and stroboscopy

## LICENSURE AND CERTIFICATION

Speech-Language Pathology Assistant License, Texas, USA, valid through 03/04/2022 Erkenning and visum logopedist, Belgium

## SCHOLARSHIP

## **PUBLICATIONS**

#### **Peer-reviewed Publications**

- Thijs, Z., Bruneel, L., De Pauw, G., Van Lierde, K. (in press). Oral myofunctional and articulation disorders in children with malocclusions: A systematic review. *Folia Phoniatrica et Logopaedica*.
- Thijs, Z., Knickerbocker, K., Watts, C. (in press). Epidemiological patterns and treatment outcomes in a private practice community voice clinic. *Journal of Voice*. https://doi.org/10.1016/j.jvoice.2020.06.025
- Thijs, Z., Watts, C. (in press). Perceptual characterization of voice quality in non-advanced stages of Parkinson's disease. *Journal of Voice*. https://doi.org/10.1016/j.jvoice.2020.05.007
- Bruneel, L., Bettens, K., De Bodt, M., D'haeseleer, E., Thijs, Z., Roche, N., & Van Lierde, K. (2020). Stages in the development and validation of a Belgian Dutch outcome tool for the perceptual evaluation of speech in patients with cleft palate. *The Cleft Palate-Craniofacial Journal*, 57(1), 43-54. https://doi.org/10.1177/1055665619862726
- Kryshtopava, M., Van Lierde, K., Defrancq, C., De Moor, M., Thijs, Z., D'haeseleer, E., Meerschman, I., Vandemaele, P., Vingerhoets, G., & Claeys, S. (2019). Brain activity during phonation in healthy female singers with supraglottic compression: An fMRI pilot study. *Logopedics Phoniatrics Vocology*, 44(3), 95-104. https://doi.org/10.1080/14015439.2017.1408853
- D'haeseleer, E., Claeys, S., Bettens, K., Leemans, L., Van Calster, A.-S., Van Damme, N., Thijs, Z., Daelman, J., Leyns, C., Van Lierde K. (2017). The impact of a teaching or singing career on the female vocal quality at the mean age of 67 years: A pilot study. *Journal of Voice, 31*(4). https://doi.org/10.1016/j.jvoice.2016.12.016

## PEER-REVIEWED CONFERENCE PRESENTATIONS

- Watts, C., Thijs, Z., Dumican, M. (2020, November). The effect of vocal intensity mode on voice production in people with Parkinson's disease [Accepted proposal]. Annual Convention of the American Speech-Language-Hearing Association, San Diego, CA (Convention canceled).
- Thijs, Z., Watts, C. (2020, May). The effect of speaking a second language on vocal function and vocal quality of life [Poster presentation]. Voice Foundation's Annual Symposium, Philadelphia, PA, USA.
- Thijs, Z., Knickerbocker, K., Watts, C. (2019, October). *Treatment outcomes in a private practice community voice clinic* [Poster presentation]. Fall Voice Conference, Plano, TX, USA.

- Thijs, Z., Watts, C. (2019, May). Perceptual assessment of phonation in speakers with Parkinson's disease [Poster presentation]. Voice Foundation's Annual Symposium, Philadelphia, PA, USA.
- Kryshtopava, M., Van Lierde, K., Defrancq, C., De Moor, M., Thijs, Z., D'Haeseleer, E., Meerschman, I., Vandemaele, P., Vingerhoets, G., Claes, S. (2018, March). *Hersenactiviteit tijdens fonatie met supraglottische compressie bij zangeressen: Een pilootstudie* [Poster presentation]. VVL-congres, Ghent, Belgium.

## **RESEARCH FUNDING**

#### **Internal Funding**

- 2020 Graduate Student Travel Grant Graduate Studies Office, Texas Christian University Travel Grant Harris College of Nursing & Health Sciences, Texas Christian University
- 2019 **Travel Grant** Harris College of Nursing & Health Sciences, Texas Christian University

#### **Honors and Awards**

- 2021 Certificate of Excellence for Outstanding Academic Achievement Fall 2020-Spring 2021, Harris College of Nursing and Health Science, TCU
- 2015-2016 Honours Programme In Life Sciences Ghent University
- 2014-2016 Honours Programme Quetelet College Ghent University

## **TEACHING EXPERIENCE**

#### **TEACHER OF RECORD**

COSD 20503 Anatomy and Physiology of the Speech and Hearing Mechanisms Texas Christian University, Davies School of Communication Sciences and Disorders Undergraduate course, taught in English Fall 2019 (face-to-face), Spring 2020 (partially face-to-face, partially online) Spring 2021 (online)

COSD 20303 Speech & Hearing Science Texas Christian University, Davies School of Communication Sciences and Disorders Undergraduate course, taught in English Fall 2020 (online)

## **OTHER SKILLS AND EXPERIENCES**

#### LANGUAGE KNOWLEDGE

**Dutch** Native language

**English** Advanced speaking, reading and writing (CEFR level C1)

**French** Conversational speaking, good reading, and writing (CEFR level B1)

#### **COMPUTER SOFTWARE KNOWLEDGE**

Technical skills include:

- Microsoft Office
- SPSS Statistics 25 (IBM)
- Praat (Boersma, P. & Weenick, D.) with Phonanium's Clinical Voice Lab (Phonanium)
- Computerized Speech Lab (Pentax Medical)

## SERVICE EXPERIENCE

- Elected senator in Texas Christian University's Graduate Student Senate for school year 2020-2021. Serving on Student Engagement & Experience Committee, and Mental Health and Wellness Committee
- Selected for **Global Outlook Institute** at Texas Christian University, a short-term, interdisciplinary global experience focusing on diversity, equity, and inclusion (attended preparatory lectures/discussions, trip canceled due to COVID-19)

## OTHER

- **Part-time education in visual arts,** certificate at primary and secondary level, specializing in drawing (2002-2018, Academie voor Beeldende Kunsten Aalst, Belgium)
- Part-time education in drama, certificate at primary and secondary level, specializing in eloquence (2005-2013, Stedelijke Academie voor Muziek, Woord en Dans, Ninove, Belgium)
- Certificate 'Animator in het jeugdwerk', training for becoming a scouts leader (2011, KSA)

#### Abstract

# SELF-PERCEIVED AFFECTIVE, BEHAVIORAL, AND COGNITIVE REACTIONS ASSOCIATED WITH VOICE AND COMMUNICATION IN PEOPLE WITH PARKINSON'S DISEASE

by Zoë Thijs, M.S., 2017 Harris College of Nursing and Health Sciences Texas Christian University

## Thesis Advisor: Christopher R. Watts, Dean of Harris College of Nursing and Health Sciences, Professor of Davies School of Communication Sciences and Disorders

**Introduction:** People with Parkinson's disease (PWPD) experience voice difficulties in the form of hypophonia. The dissertation aimed to describe the psychosocial consequences of this disorder in PWPD. More specifically, it intended to describe the affective, cognitive, and behavioral reactions to the voice disorders experienced by PWPD.

**Method:** PWPD and healthy controls completed the Behavior Assessment Battery-Voice (BAB-Voice). Daily communications partners of both groups completed an adapted version of the BAB-Voice. The instrument was administered via clinician-guided or participant-guided mode. Subtest scores were calculated and analyzed.

**Results:** There was no difference in administration mode. PWPD scored significantly higher than healthy controls, indicating increased psychosocial burden due to hypophonia. Communication partners' ratings agreed with the PWPD's own ratings.

**Conclusions:** Compared to healthy controls, PWPD experience more negative affective, behavioral, and cognitive reactions to voice. Moreover, daily communication partners can provide valuable information for diagnosis and treatment of the voice disorder in PD.