Self-Perceived Affective, Behavioral and Cognitive Reactions Associated with Voice Use in People with Parkinson's Disease: A Pilot study

Zoë Thijs^a*, Yan Zhang^a, Kristiane Van Lierde^{b, c}, Martine Vanryckeghem^d, Christopher Watts^a

^a Harris College of Nursing and Health Sciences, Texas Christian University, Fort Worth, Texas, USA; ^bCenter of Speech and Language Sciences, Department of Rehabilitation Sciences, Ghent University, Ghent, Belgium; ^cDepartment of Speech-Language Pathology and Audiology, University of Pretoria, Pretoria, South-Africa; ^dSchool of Communication Sciences and Disorders, University of Central Florida, Orlando, Florida, USA

* Zoë Thijs, PhD
Harris College of Nursing & Health Sciences
Texas Christian University
TCU Box 298625
Fort Worth, TX 76129
zoe.thijs@tcu.edu

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Purpose: This study aimed to compare the affective, behavioral, and cognitive reactions related to vocal function in People with Parkinson's Disease (PWPD) and healthy controls using the Behavior Assessment Battery – Voice (BAB-Voice). The tests' internal consistency was also described.

Methods: 31 PWPD and 19 healthy controls were recruited from September 2020 to March 2021. Participants completed four BAB-Voice subtests: Speech Situation Checklist - Emotional Reaction (SSC-ER), the Speech Situation Checklist - Speech Disruption (SSC-SD), Behavior Checklist (BCL), and Communication Attitude Test for Adults (BigCAT), describing the experienced negative emotional reaction, voice disruptions, coping behaviors, and negative attitude regarding communication respectively. Subtest scores were calculated and analyzed.

Results: The scores of the PWPD were significantly different from those of the controls (Pillai's Trace=0.344, F[4]=5.508, p=0.001, η_p^2 =0.344): PWPD showed more negative emotions and voice problems, more coping behaviors, and more negative speech-related attitude compared to healthy controls. All subtests showed excellent internal consistency.

Conclusions: The BAB-Voice proved a tool with a good internal consistency that measured different psychosocial reactions in PWPD versus controls. PWPD exhibited significantly more negative emotions and voice problems in specific speech situations, more coping behaviors, and more negative speech-related attitude. The specificity of information obtained from the BAB-Voice may aid in improving the treatment planning of voice disorders in PWPD.

Keywords: Parkinson's disease; voice disorder; hypophonia; psychosocial effect; Behavior Assessment Battery

Word count: 3459

Introduction

Up to 90% of people with Parkinson's disease (PWPD) will exhibit speech impairments during their disease process [1,2]. The hypokinetic dysarthria in PWPD affects multiple domains of speech, for example, disordered articulatory movements and disturbances of speech rate. Specifically for the phonatory system, low-intensity voice along with a harsh and breathy voice quality is apparent, in addition to prosodic disturbances and possibly a vocal tremor [1,3,4]. These voice problems are often collectively referred to as hypophonia [4,5].

Hypokinetic dysarthria and hypophonia influence the psychosocial wellbeing of PWPD. They experience negative emotions, such as frustration and embarrassment, when communicating [6,7]. Moreover, PWPD think more negatively about their communicative ability. They might feel less confident, less in control, and less independent when speaking [6]. To deal with the communicative difficulty and its consequences, PWPD may utilize coping behaviors [7]. Some of these strategies include increasing physical effort when speaking, reducing or even avoiding conversations, or informing their environment of the experienced communicative difficulty [2,7,8]. As a result, PWPD may experience social withdrawal and participate less in multiple aspects of daily living, such as work-life and leisure life [2,7,8].

Several patient-reported outcome measures focusing on voice disorders have been developed over the years [9,10]. These instruments capture the patient experience of the vocal symptoms and/or the impact it has on their daily lives [9,11], and thus could capture the above described psychosocial impact. Many of these self-assessment tools fit within a multidimensional framework as they capture information additional to what a clinician can measure during the voice evaluation. One such system is the International Classification of Functioning, Disability, and Health (ICF), a multidimensional framework developed to measure disability [12,13]. The ICF considers disability as the interaction of the health condition with the environment. It describes both "Functioning and disability" (i.e., "Body function", "body structure", "activities", "participation") and "Contextual factors" (i.e., "Environmental factors", "Personal factors") and how they interact with one another [12,13]. The ICF framework has been considered an appropriate tool to use in speech-language pathology practice to promote person-centered approaches and was included in the Scope of Practice for Speech-Language Pathology and Audiology [14].

A wide variety of patient-reported outcome measures has been used in the PD population [15–22], but they present some challenges. The majority of the patientreported outcome measures focusing on the psychosocial consequences center around the "participation" level in the ICF model (e.g., Dysarthria Impact Profile, DIP [23]; Communicative Effectiveness Survey, CES [24]; Communication Participation Item Bank, CPIB [25,26]) or "activity" and "participation" level (e.g., Voice Activity and Participation Profile, VAPP [27]). Other tools provide more general information on multiple factors within the ICF model but have a limited number of questions (e.g., Voice Handicap Index, VHI [28]; Voice-Related Quality of Life, V-RQOL [29]), or are not standardized and normed (e.g, DIP [23], VAPP [27]). However, a detailed description of the affective, behavioral, and cognitive reactions to voice use and their situational differences could potentially inform clinical treatment planning through identifying specific feelings and speech production during challenging situations, and the use of coping behaviors and negative speech-related thinking which might need to be addressed during therapy. Moreover, this information could provide important, not only in light of diagnosis and treatment planning but also in light of peri- and posttreatment outcomes. Consequently, a more extensive tool is a useful addition to the current patient-reported outcome measures.

In communication disorders other than dysphonia, assessing psychosocial consequences in detail and incorporating that information into treatment planning is common practice. In people who stutter (PWS), the Behavior Assessment Battery (BAB) [30] has been shown to be a useful standardized and normed assessment tool in terms of differential diagnosis, which also serves as a road map indicating the treatment targets, and as an outcome measure. The BAB assesses the presence of affective, behavioral, and cognitive reactions to communication by means of self-report [30]. It has also been adapted to a population with a neurological voice disorder, spasmodic dysphonia (SD), the BAB-Voice [31–33]. The use of this tool has found significant between-group differences for participants with SD and controls for all BAB sub-tests [31].

The BAB-Voice consists of four different subtests looking at different psychosocial aspects related to voice use. The Speech Situation Checklist – Emotional Reaction (SSC-ER) and the Speech Situation Checklist – Speech Disruption (SSC-SD) judge how different speech situations affect negative emotions such as anxiety, fear and worry, and the experienced voice problems, respectively. These two subtests can be framed within the "activities", "participation", and "personal factors" of the ICF model. The Behavior Checklist (BCL) considers the possible use of coping behaviors, which are "personal factors". The Communication Attitude Test for Adults (BigCAT) addresses the speaker's voice-related beliefs and attitude, once again considering multiple aspects of the ICF model: "activities", "participation", "personal factors", "environment" [31–33]. Using the BAB-Voice to establish normative data, it has been shown that individuals with SD present statistically significantly more negative feelings (e.g., anxiety), more coping behaviors, and more negative attitude than a control population [31–33]. Considering the usefulness of the tool in a voice-disordered population, the BAB-Voice could potentially be useful in providing more information on the psychosocial consequences of hypophonia in PWPD.

Given that the psychosocial effect that PWPD experience due to hypophonia remains unclear, the current study aimed to compare the self-perceived affective, behavioral, and cognitive reactions to hypophonia in PWPD and healthy controls. As this study was the first to administer the BAB-Voice to PWPD, the relationship between the BAB-Voice subtests and internal consistency was also considered. The specific questions addressed were: (1) What are the affective, behavioral, and cognitive reactions to voice use in PWPD and healthy controls, (2) What is the internal consistency of the BAB-Voice-items within the different subtests when using it with PWPD? The following hypotheses were considered: (1) the affective, behavioral, and cognitive reactions to voice would be more negative in PWPD compared to healthy controls, and (2) based on previous reports of the BAB and BAB-voice in different populations, the internal consistency of the BAB-Voice would be strong.

Material and Methods

Population

The study was approved by the first author's Institutional Review Board. PWPD and healthy controls were recruited for the study. To participate, PWPD had to be diagnosed with PD by a neurologist, self-report hearing to be within normal limits for their age with or without hearing amplification, and have no additional diagnosed neurological issues other than PD. Outside of their experience with PD, participants could not have a history of other speech, voice, or language disorders. PWPD were recruited in the United States and contacted through an existing volunteer database, local PD support groups, as well as social media from September 2020 to March 2021. Healthy controls were also required to self-report normal hearing for their age and could not have any diagnosed neurological disorders or a history of speech, voice, or language disorders. Controls were enlisted through senior groups and social media, as well as outreach in the local community.

Data Tools

All participants completed a demographic questionnaire as well as an adapted version of the BAB-Voice with its four subtests. The demographic questionnaire asked about general information (biological sex, age), details about Parkinson's disease (onset, treatment history), and inclusion/exclusion criteria (presence of other neurological diagnoses, self-reported hearing status, history of speech, voice, or language therapy).

Both the SSC-ER and SSC-SD subtests consist of 38 different speech situations (e.g., "talking on the phone", "talking to a stranger"). The SSC-ER asks participants to rate the negative emotional reactions (anxiety, fear, worry) experienced when speaking in those specific situations on a scale from 0 ("not at all") to 5 ("very much"), whereas the SSC-SD uses an identical scale to rate the experienced voice problems for those same situations. Both subtests thus can result in a score between 38 and 190, with a higher score indicating more experienced negative emotional reactions or voice problems. The BCL subtest consists of a description of 34 behaviors that a person can use to cope with voice problems. A "no" answer indicates that the participant reports not to use the coping behavior and is scored 0. A "yes" response confirms that the participant does use the particular coping behavior and is scored 1. As such, the resulting score can be between 0 - 34, with a higher score indicating more coping behaviors. The subtest items include coping behaviors such as "avoiding eye contact" or "taking a deep breath before speaking". The BigCAT subtest comprises 34 true/false statements about voice-related attitude, how the participant currently thinks about their voice. This dichotomous "True-False" scale is rated either 0 or 1, with 1 indicating a negative attitude. For example, a "True" response to the item "There is something wrong with my voice" would be scored as 1. This subtest may result in a score between 0 and 34, with a higher score indicating a more negative attitude.

Data collection

Data were collected between September 2020 and March 2021 when direct human contact was limited due to the COVID pandemic. Participants were invited to fill out the questionnaire virtually, either independently via Qualtrics or with the support of a researcher while utilizing Qualtrics via Zoom. After informed consent, the participants filled out the demographic questionnaire. Next, a brief description of normal and dysphonic voice, as well as instructions that the following questions related to situations where voice would be used and perceived by others (e.g., the focus of the questions were on voice, not the larger construct of speech), was shown. Participants were required to indicate that they understood the voice-related focus of the questionnaire. A reminder to consider voice was also added to the instructions of each BAB-Voice subtest. The participants then completed the BAB-Voice subtests in a randomized order. Due to institutional restrictions related to COVID-19 during the data collection, no associated voice recordings could be collected.

Statistical Analysis

The data were first analyzed descriptively and visually using SPSS (version 25.0, Armonk, NY: IBM Corp). All analyses employed a significance level of α =0.05. Outliers falling outside of 3x interquartile range were removed prior to the analysis as these extreme values likely do not represent the target population, resulting in the removal of 2 data points. Demographic data were compared between PWPD and control groups using Mann-Whitney U tests for the continuous variables (i.e., chronological age) due to non-normality, and chi-square tests for the categorical variables (i.e., sex). The differences of BAB-Voice scores between the disease status (PWPD vs. healthy controls) while controlling for age were compared using a MANCOVA. Disease status was the independent variable, the scores of all four subtests were the dependent variables, and age was incorporated as the covariate. Follow-up analysis with Bonferroni correction was performed as needed. The internal consistency of the BAB-Voice was considered using two-way mixed, absolute agreement, average measures intraclass correlation coefficient (ICC) for each of the subtests.

Results

The study included 50 participants (31 PWPD, 19 controls). Demographic characteristics of the overall sample can be found in Table 1. A statistically significant difference for chronological age was found between the PWPD and controls (U=455.0, p=0.001). The PWPD were on average older (M=71.23 years, SD=9.09) compared to the healthy controls (M=63.79 years, SD=7.04). Therefore, age was controlled in the multivariate analysis as a covariate. The scores for each of the subtests are described in Table 2. Numerically, the PWPD showed higher scores than healthy controls on all four subtests of the BAB-Voice, indicating more negative affective, behavioral, and cognitive reactions to communication.

		PWPD (n=31)	Control (n=19)	Test result
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
Chronological	Mean (SD)	71.23 (9.09)	63.79 (7.04)	<i>U</i> =455.0,
Age (years)	Median (IQR)	73 (65-78)	62 (59-69)	<i>p</i> =0.001
PD duration	Mean (SD)	8.02 (5.13)		
(years)	Median (IQR)	7.33 (4-10.75)		
PD treatment	Treated	31 (100%)		
	- Medication	- 30		
	- DBS	(96.8%)		
	- Other	- 6(19.4%)		
		- 7 (22.6%)		
SLT in past	SLT in past	12 (38.7%)		
	SLT currently	2 (6.5%)		

Table 1. The descriptive data of the overall sample of PWPD and healthy controls

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; PD, Parkinson's disease; SLT, speech-language therapy; SD, standard deviation; IQR, interquartile range; DBS, deep brain stimulation.

Table 2. The BAB-Voice subtest scores of the overall sample of PWPD and healthy controls

		PWPD (n=31)	Control (n=19)
SSC-ER	Mean (SD)	82.97 (46.75)	48.89 (11.35)
	Median (IQR)	60 (50-124)	46 (38-55)
SSC-SD	Mean (SD)	77.32 (40.17)	45.68 (9.24)
	Median (IQR)	65 (48-96)	42 (39-51)
BCL	Mean (SD)	7.19 (5.84)	2.68 (3.99)
	Median (IQR)	7 (2-9)	1 (0-4)
BigCAT	Mean (SD)	16.32 (11.71)	2.29 (2.26)
	Median (IQR)	13 (6-27)	2 (1-4)

Note. The average and median scores for each subtest are represented for both groups. The pairwise comparison was calculated as a follow-up analysis of the MANCOVA analysis.

Abbreviations. PWPD, people with Parkinson's Disease; 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.

A MANCOVA was performed to compare disease status (PD vs. control) for the four subtest scores while controlling for age. No significant effect was found for the covariate age (Pillai's Trace=0.027, F[4]=0.276, p=0.891, η_p^2 =0.027). There was a significant effect of disease status (Pillai's Trace=0.344, F[4]=5.508, p=0.001, η_p^2 =0.344, Observed Power=0.961) on the combined BAB-voice four subtest scores. One-way ANOVA follow-up tests revealed that PWPD scored statistically significantly higher than the controls for all subtests with the observed power being near or above 80%. This indicated the presence of significantly more negative emotional reactions in speech situations (SSC-ER, F[1]=8.830, p=0.005, η_p^2 =0.164, Observed Power=0.828), more experienced voice problems in those same situations (F[1]=9.387, p=0.004, η_p^2 =0.173, Observed Power=0.850), more coping behaviors (BCL, F[1]=6.722, p=0.013, η_p^2 =0.130, Observed Power=0.718), and a more negative voice-related attitude (BigCAT, F[1]=21.342, p<0.001, η_p^2 =0.322, Observed Power=0.850) (see Table 2 and Figures 1-4) among PWPD than the health controls. The biggest difference in the BAB-voice scores is shown in the voice-related attitude (BigCAT), with PWPD scoring

(16.32 ±11.71) scoring six standard deviations (SD) above the healthy controls (2.29±2.26). PWPD scored three SD above the healthy controls when comparing the negative emotions (SSC-ER, PWPD: 82.97±46.75, controls: 48.89±11.35) and voice problems in specific speech situations (SSC-SD, PWPD: 77.32±40.17, controls: 45.68±9.24). The smallest difference in terms of SD was found for the coping behaviors (BCL): PWPD (7.19±5.84) scored 1 SD higher than the controls (2.68±3.99).

Based on Cicchetti's [34] criteria, the two-way mixed, consistency, average measures ICC revealed that the items of the BCL were in excellent agreement with one another. The BigCAT, SSC-ER, and SSC-SD showed similar results (see Table 3).

	Intraclass	95% Confidence	Judgment based
	Correlation	interval	on Cicchetti (34)
SSC-ER	0.992	0.988-0.995	Excellent
SSC-SD	0.991	0.986-0.994	Excellent
BCL	0.882	0.830-0.924	Excellent
BigCAT	0.970	0.956-0.980	Excellent

Table 3. The 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

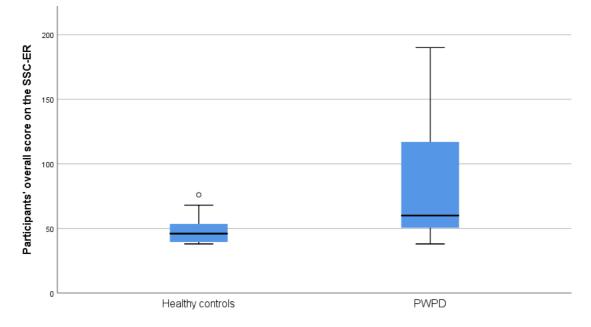


Figure 1. Self-rated scores of PWPD and healthy adults on the SSC-ER

Abbreviations. SSC-ER, Speech Situation Checklist – Emotional Reaction; PWPD, people with Parkinson's disease.

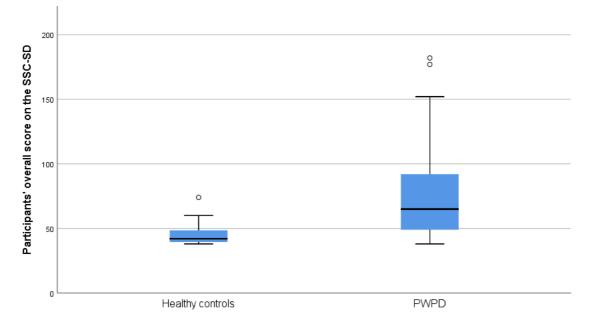


Figure 2. Self-rated scores of PWPD and healthy adults on the SSC-SD

Abbreviations. SSC-SD, Speech Situation Checklist – Speech Disruption; PWPD, people with Parkinson's disease.

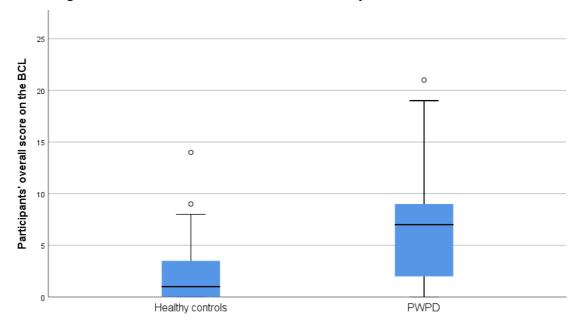


Figure 3. Self-rated scores of PWPD and healthy adults on the BCL

Abbreviations. BCL, Behavior Checklist; PWPD, people with Parkinson's disease.

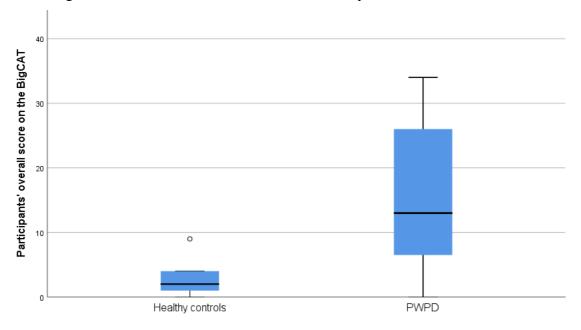


Figure 4. Self-rated scores of PWPD and healthy adults on the CAT

Abbreviations. CAT, Communication Attitude Test for Adults; PWPD, people with Parkinson's disease.

Discussion

The current study aimed to determine the difference in affective, behavioral, and cognitive reactions related to voice use between PWPD and adults without PD (controls) using the BAB-Voice self-report tests as the method of assessment. The study found that PWPD exhibited statistically significantly more negative affective, behavioral, and cognitive reactions compared to healthy adults, as evidenced by significantly higher scores on the BAB-Voice.

Our findings agree with existing reports. Previous studies have established substantial psychosocial effects due to communication impairment associated with PD. As previously mentioned, these negative effects can include frustration or embarrassment, difficulty in specific speech situations, and the presence of coping behaviors [2,7,35,36]. In the current study, these behaviors were quantified in communicative situations with a focus on voice-related reactions and behaviors. Our results were in agreement with what other measurement tools have described in a population with PD. Letanneux et al. [17] and Cardoso et al. [37] found that the DIP was able to discriminate between PWPD and controls. The PWPD presented with statistically significantly lower scores on the DIP, which indicated a larger psychosocial impact. Similarly, for the CES, Donovan et al. [18] and Dykstra et al. [38] found statistically significantly lower scores for the PWPD compared to controls, indicating less perceived effectiveness during communication. The voice-specific measures, VHI and V-RQOL, also found a significant difference between PWPD and controls, with the scores of PWPD indicating worse voice-related quality of life [20,21]. Along with findings from the present study, the evidence collectively suggests that there is a significant psychosocial burden associated with the speech and voice deficits experienced in PD. Moreover, this burden associated with voice impairment can be measured specifically using the BAB-Voice.

Interestingly, a large degree of variability was present in the subtest scores of the BAB-Voice, especially in the PWPD group. This could be explained by the clinical heterogeneity within the presentation of PD [39]. More specifically, PWPD tend to present with a great variety of vocal and speech symptoms throughout their disease process [40,41]. The variability within our results could therefore be due to our recruitment strategy: the presence of a diagnosed voice disorder was not a prerequisite to participate in the study. Given the range of time since onset of PD within our sample

(between less than a year to over 22 years), we likely recruited PWPD representing a wide range of disease progression. Another possible explanation is that PWPD are not always aware of their voice impairments, as perceptual deficits have been reported in PWPD [42]. For example, it has been described that PWPD do not accurately perceive their vocal intensity [41,43,44]. Therefore, PWPD potentially underestimated their vocal complaints, which would influence the related psychosocial reactions. Despite large inter-individual differences, strong internal consistency was found for each subtest. This finding was similar to the previous BAB-Voice reports [31] and indicated that the items on the subtests likely measured one construct for each subtest.

The BAB has originally been administrated to populations of PWS and, later on, to those with spasmodic dysphonia (SD). Therefore, subtest scores from the current study could be compared to extant data. SD is a focal neuropathology restricted to the level of the larynx [45]. While both SD and PD are neurological disorders leading to changes in voice physiology, the focal and non-degenerative nature of SD distinguishes both. BAB-Voice scores of people with SD were significantly higher than those of healthy controls [31]. Moreover, the scores were distributed towards the extreme end of the scale for all subtests, indicating a high amount of negative emotional reactions (SSD-ER), speech disruption (SSC-SD) in different speaking situations, frequent use of coping behaviors (BCL), and a very negative speech-related attitude (BigCAT) [31–33]. PWPD overall scored lower than those with SD, with scores in the mid-to-low ranges of each subtest. These differences could be explained by the different nature of speech disorders. Stuttering and SD are disorders that suddenly and transiently impact the normal flow of speech and present with similar symptoms [31,32,46,47]. Both disorders present with dysfluent speech, with typically more impact on meaningful or complex speech [47–49]. SD has even been called "laryngeal stutter" [47]. While basal ganglia dysfunction has been suggested in stuttering, SD, and PD [46,50], the changes present in PD are more gradual. They are continuously present during speech production [1,44,51]. Though interruptions and dysfluencies have been reported in PD, these are not the most common or obvious characteristics of hypokinetic dysarthria. The sudden and transient changes during speech in SD and stuttering may be perceived as more salient and impairing than the gradual changes developed later in life seen in PD.

One potential advantage of the BAB-Voice is that it provides an extensive and holistic view of a person's responses to communication situations associated with voice production [31], which has potential as a diagnostic and outcome measure and a tool to

inform treatment. The ICF model can be used to guide treatment and assess its effects [12,13]. Including psychosocial measures post-therapy is therefore recommended and often implemented in practice. The BAB-Voice could serve as an addition to the more commonly used and more succinct measures such as the VHI. The detailed information gathered by the assessment tool (e.g. differences in how challenging speech situations are perceived, the use of specific coping behaviors, a negative speech-associated attitude) can additionally inform a multimodal and holistic approach to treatment planning. Given that the psychosocial impact is not always treated in patients with hypokinetic dysarthria [52], the specificity of the BAB-Voice could facilitate the identification of novel and impactful treatment targets [31]. More research is needed to determine the value of more holistic treatment approaches in PWPD.

Some limitations were manifested in the current study. The PWPD and control group sizes were small and unbalanced. The presence of COVID-19 during the time of recruitment could be a possible explanation for this. Moreover, given the older population that was recruited in this study, some participants may not have been comfortable filling out a survey digitally. The length of the BAB-Voice instrument may also have contributed to incomplete responses and denial of prospective participants to participate. Allowing more flexible administration of the BAB-Voice (e.g. by allowing multiple days to fill out the instrument) or exploring shortening the instrument without compromising its validity and reliability could potentially increase the sample size of future projects and the feasibility of administration to the PD population. However, despite the small sample size, our analyses showed adequate power. The current study focused primarily on voice use. It may not have been easy for participants to single out this subsystem of speech, given that hypokinetic dysarthria affects all subsystems [1,3,4]. Future research could compare results on the BAB when focusing solely on those with diagnosed hypophonia versus those without. Finally, the current project could not compare the perceived psychosocial impact of voice use with auditoryperceptual, acoustic, or laryngeal videostroboscopic consensus evaluation of voice production, as COVID-19-regulations during the time of the study limited data collection options. Future research can include these measures to see if relationships exist with the experienced psychosocial impact.

Conclusion

This study aimed to compare the psychosocial reactions of PWDP and controls. Based on the subtest scores from the BAB-Voice, PWPD exhibited significantly more experienced negative emotions and voice problems in specific speech situations, more coping behaviors, and more negative speech-related attitude. These results were in accordance with previous literature, indicating that PWPD experience a significant psychosocial impact due to their hypophonia. Based on these findings, speech-language pathologists should consider treating the psychosocial consequences of the communication disorder as well as the hypophonia itself. More research is needed on these multidimensional treatment approaches.

Declaration of interest

The fourth author (M.V.) has a personal, non-financial interest as test author of the BAB-Voice. The other authors report no declarations of interest.

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