

Acquired Stuttering in Parkinson's Disease

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Abstract: Background: Parkinson's disease frequently causes communication impairments, but knowledge about the occurrence of new-onset stuttering is limited.

Objectives: To determine the presence of acquired neurogenic stuttering and its relationship with cognitive and motor functioning in individuals with Parkinson's disease.

Method: Conversation, picture description, and reading samples were collected from 100 people with Parkinson's disease and 25 controls to identify the presence of stuttered disfluencies (SD) and their association with neuropsychological test performance and motor function.

Results: Participants with Parkinson's disease presented with twice as many stuttered disfluencies during conversation ($2.2\% \pm 1.8\%SD$) compared to control participants ($1.2\% \pm 1.2\%SD$; $P < 0.01$). 21% of people with Parkinson's disease ($n = 20/94$) met the diagnostic criterion for stuttering, compared with 1/25 controls. Stuttered disfluencies also differed significantly across speech tasks, with more disfluencies during conversation compared to reading ($P < 0.01$). Stuttered disfluencies in those with Parkinson's disease were associated with longer time since disease onset ($P < 0.01$), higher levodopa equivalent dosage ($P < 0.01$), and lower cognitive ($P < 0.01$) and motor scores ($P < 0.01$).

Conclusion: One in five participants with Parkinson's disease presented with acquired neurogenic stuttering, suggesting that speech disfluency assessment, monitoring and intervention should be part of standard care. Conversation was the most informative task for identifying stuttered disfluencies. The frequency of stuttered disfluencies was higher in participants with worse motor functioning, and lower cognitive functioning. This challenges previous suggestions that the development of stuttered disfluencies in Parkinson's disease has purely a motoric basis.

People with Parkinson's disease (PD) commonly develop communication disorders. Up to 90% will present with changes in voice and articulatory precision.¹ Emerging evidence suggests that PD can also be associated with the development of acquired neurogenic stuttering.²⁻⁴

Acquired neurogenic stuttering occurs following neurological injury or disease, in people with a history of fluent speech production. This contrasts with developmental stuttering, which emerges during child development.⁵ The core characteristic in both cases of developmental and acquired stuttering is the occurrence of stuttered disfluencies (SD), ie, part-word repetitions (eg, "t-t-t-train"), mono-syllabic word repetitions (eg, the-the-the-

the cat), prolongations (eg, "ssssand"), and blocks (eg, "... car").^{6,7} These need to be distinguished from typical disfluencies present in normal speech, such as phrase repetitions (eg, "I walked, I walked to the shops"); revisions (eg, "I walked to the café, to the shops"); incomplete phrases (eg, "I went to ..."); interjections (eg, "I um love uh apples") and word finding pauses (eg, "I went to the ... cinema").^{6,7}

The pathophysiologic mechanism underlying acquired neurogenic stuttering is currently not well understood.⁸ In patients with stroke-induced stuttering, brain lesions do not appear to be limited to one specific brain area,^{9,10} but have been associated with lesions in the left-sided cortico-basal ganglia-cortical

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network for speech production.¹¹ Involvement of the basal ganglia circuits has also been associated with onset of acquired neurogenic stuttering following traumatic brain injury and drug use.^{11–14} In addition, impairment to the basal ganglia-thalamocortical circuit and associated dopaminergic changes have been suggested as possible neural causes of stuttering.^{15,16} In computational modeling of speech production, both low and high levels of dopamine impact on the model's basal ganglia-thalamus-left ventral premotor cortex circuit can result in stuttering moments.¹⁷

Further support for the involvement of a dopaminergic influence in stuttering comes from cases with stuttering onset in progressive supranuclear palsy and manganese-induced ephedrone parkinsonism.^{18–20} As PD is associated with dopaminergic and consequently basal ganglia degeneration,²¹ studying the occurrence of acquired neurogenic stuttering in this population may help to elucidate the pathophysiological mechanisms underpinning the occurrence of stuttered disfluencies.

In PD, degeneration of the dopaminergic neurons of the substantia nigra triggers functional re-arrangement and changes to the entire basal ganglia network.²¹ Initially, the neuronal damage and loss mostly occurs in areas involved in motor function. However, as the disease progresses there is wider cerebral involvement, reflected by changes to cognition in particular.^{22,23} To ensure adequate follow-up and care is provided for those with PD, it is important to understand the full range of changes that can affect quality of life, including the development of stuttered disfluencies.²⁴

Only two previous studies directly compared stuttered disfluencies in the speech of people with PD and healthy older adults, revealing a higher frequency of stuttering disfluencies in the PD group ($n = 32$ and 20 , in Goberman et al²⁵ and Juste et al²⁶ respectively). Their analyses were restricted to monologue and reading tasks. Variability across different speech tasks—with highest occurrence of stuttering during conversation—is well established for developmental stuttering and has also been reported in stuttering following stroke and TBI, but has not been examined in PD.^{26–28} It is important to assess the presence of stuttered disfluencies during conversation, as this most closely matches everyday communication.

In addition, the link between the occurrence of stuttered disfluencies and features of disease progression is currently unclear. Previous studies, not including a healthy control group, have focused on the relationship between stuttered disfluencies and medication as well as disease duration. One study of 51 participants with PD in both on and off states of medication showed no significant relationship between the severity of speech disfluency in a reading task and disease duration or levodopa equivalent dosage.²⁹ However, another study of 14 people with PD found a significant correlation between higher cumulative dosages of dopaminergic medication and a higher frequency of stuttered disfluencies.⁴ It is unclear whether this correlation reflects a direct causal relationship or represents a more complex relationship mediated by factors such as response to medication and symptom severity.⁴

Traditionally, speech changes such as dysarthria in PD have been attributed to motor deficits such as bradykinesia and rigidity.^{30–32} Consistent with this view, correlations between speech disfluencies and freezing of gait have been reported,³³ leading researchers to suggest that the speech disfluencies in PD are also motor based.^{25,26} However, cognitive contributions to speech changes are increasingly being recognized, and understanding whether cognitive factors additionally contribute to stuttered disfluencies is important for both theoretical (eg, understanding causal mechanisms) and clinical (eg, treatment focus) purposes.^{4,31,32,34,35}

To address these needs, we will directly compare the occurrence of stuttered disfluencies during conversation in a large group of PD participants and controls. Next, by measuring stuttered disfluencies across speech tasks, we aim to identify task-related differences and establish the preferred speech task for identification of stuttering in PD. Finally, we will investigate the link between stuttered disfluencies and cognitive and motor measures of disease progression. In addition to assessing the influence of each of these factors separately, we are specifically interested in whether reduced cognitive function leads to a further increase in stuttered disfluencies beyond changes attributable to motor symptoms. Together, these findings can lead to improvements in identification, monitoring, and treatment of stuttering in PD.

Methods

Participants

A convenience sample of 103 PD participants was recruited from our research institute and movement disorders clinic. A total of 27 age-similar healthy controls who responded to community advertisements were included in the study.^{36,37} Participants gave written informed consent, and the study was approved by the New Zealand Health and Disability Ethics Committee (URB/09/08/037/AM22). Four participants (2 PD, 2 Control) were excluded due to a history of developmental stuttering and one (PD) because of a subsequent diagnosis of progressive supranuclear palsy, leading to a final sample of 100 PD participants (62 Male, 38 Female) and 25 controls (13 Male, 12 Female). Demographic information including age, education level, disease and symptom onset age, handedness, side of onset, and Hoehn and Yahr overall disease severity scores,³⁸ are displayed in Table 1. All assessment sessions occurred before 12 pm to minimize variations due to fatigue and medication on/off periods. Nearly all (92/100) PD participants were taking a range of Parkinson's medications (quantified as Levodopa Equivalent Dosage [LED]) at the time of assessment. Based on self-report, 90 participants reported that their medication was currently working (on-state) and two felt their medication was not currently working (off-state). Three participants in the on-state also reported receiving deep brain stimulation therapy. One participant reported receiving deep brain stimulation and was not taking medication at the time of assessment.

TABLE 1 Demographic information

Demographic	PD (<i>n</i> = 100)	Control (<i>n</i> = 25)	<i>P</i> -value (Wilcoxon value)	Effect size—Cohen's <i>d</i>
Biological sex				
Male/female	62/38	13/12	—	—
Age				
Mean (standard deviation)	72 (7.0)	76 (6.9)	0.01 (1656)	0.50
Education				
Mean (standard deviation)	13 (2.6)	13 (2.7)	0.43 (1376)	0.17
Diagnosis age				
Mean (standard deviation)	62 (8.6)	—	—	—
Years since diagnosis				
Mean (standard deviation)	10 (0.6)	—	—	—
Symptom onset age				
Mean (standard deviation)	60 (8.9)	—	—	—
Years since symptom onset				
Mean (standard deviation)	12 (6.7)	—	—	—
Side of onset				
Right/left/bilateral	48/46/4	—	—	—
Levodopa equivalent dosage				
Mean (standard deviation)	841 (553)	—	—	—
Handedness				
Right/left/ambidextrous	91/7/1	21/1/1	—	—
Hoehn & Yahr				
Median (range)	2.5 (1–4)	—	—	—

Cognitive and Motor Assessment

All participants completed a comprehensive neuropsychological battery, consisting of assessments in five cognitive domains: attention, working memory and processing speed; executive function; visuoperceptual/visuospatial; learning and memory; and language (Table S1).^{37,39} The battery meets the Movement Disorder Society-task force's Level II PD-Mild cognitive impairment (PD-MCI) criteria (at least two neuropsychological tests in each of the five cognitive domains).^{37,40} Participants were classified as PD-MCI if they had two impaired scores (<1.5 standard deviations below the normative data average) in a single domain.^{37,40} PD-Dementia (PDD) was classified as two or more impairments (<2.0 standard deviations below the normative data average) in at least two of the five cognitive domains as well as functional impairment to their daily activities.⁴¹ Those not meeting either of the criteria for cognitive impairment were classified as PD with normal cognition (PD-N). A global Z score (expressed as an aggregated Z score), an estimation of global cognitive ability, was derived by averaging performance across the 22 neuropsychological variables assessed (Table S1).

Motor function was assessed in participants with PD (unavailable for six), using Part III of the Movement Disorder Society-Unified Parkinson's Disease Rating Scale (MDS-UPDRS).⁴²

Speech Samples

Speech samples were collected during three speech tasks. Conversations with the examiner started by discussing family and hobbies, and expanding to interests, travel, etc. as required to elicit speech for approximately 10 min. Conversation involved turn taking between the examiner and the participant with both parties able to ask questions and contribute to the conversation. Verbal prompts (eg, follow-up questions, asking for more detail) were used to extend the conversational discourse as required to ensure samples were of an adequate length (>450 words). For the picture description task participants were asked to describe the "Cookie Theft"⁴³ picture, by telling a story using full sentences. If needed, verbal prompts (eg, "why do you think the mother is distracted?," "what do you think will happen when the mother turns around?") were used so that the picture description included at least 150 words. For the third task,

participants were asked to read aloud “The Caterpillar”⁴⁴ passage (197 words) at a comfortable, habitual pace. Speech tasks were video recorded with the participant’s head, shoulders, and torso in view, on a MacBook Air laptop computer using the multimedia framework QuickTime⁴⁵ and a Logitech C615 webcam (maximum 1920 × 1080 pixel resolution and 30fps frame rate with one built in omni-directional noise-reducing microphone). One participant requested audio-only recordings. Six conversation samples, 22 picture description samples and one reading sample from a total of 22 participants (21 PD, 1 Control) were too short and were therefore excluded from the analyses.

The first 450 words of the participants’ conversation samples and first 150 words of the participants’ picture description samples were transcribed verbatim (with a ± 10-word margin to end on a complete utterance). The full reading samples were transcribed. All transcriptions were done by the first author in Computerized Language ANalysis (CLAN) software, coding speech disfluencies per the CLAN manual.⁴⁶ Due to the use of video recordings, the transcriber was not blinded to all participants’ diagnoses. Data on total syllables and stuttered disfluencies (number of prolongations, blocks, part-word repetitions, and mono-syllabic word repetitions) was extracted from CLAN and the frequency of stuttered disfluencies per 100 syllables was calculated ($\%SD = \left(\frac{\text{number of stuttered disfluencies}}{\text{total number of syllables}} \right) \times 100$).⁴⁷ Consistent with previous literature in acquired stuttering following stroke and traumatic brain injury,^{28,48} the commonly utilized criterion of ≥3% stuttered syllables during conversation was the threshold used for a stuttering diagnosis.

Intra-rater reliability was calculated by rescored 17% of conversation, monologue and reading transcripts by the first author after a delay of at least 10 months, using Pearson’s correlation measure (conversation, $r = 0.98$, $P < 0.01$; picture description, $r = 0.97$, $P < 0.01$; and reading tasks, $r = 0.79$, $P < 0.01$). Similarly strong intra-rater reliability was found using interclass correlation coefficient for conversation (ICC = 0.99, $P < 0.01$), monologue (ICC = 0.98, $P < 0.01$), and reading (ICC = 0.87, $P < 0.01$). Inter-rater reliability was calculated by rescored 17% of conversation, monologue and reading transcripts by a qualified Speech and Language Therapist with experience scoring stuttering, using Pearson’s correlation measure (conversation, $r = 0.93$, $P < 0.01$; picture description, $r = 0.93$, $P < 0.01$; and reading tasks, $r = 0.87$, $P < 0.01$). Excellent inter-rater reliability was also found using interclass correlation coefficient for conversation (ICC = 0.96, $P < 0.01$), monologue (ICC = 0.96, $P < 0.01$), and reading (ICC = 0.93, $P < 0.01$).

Statistical Analysis

Group differences for age, years of education, and global cognitive functioning were assessed using Wilcoxon rank-sum tests (W), with effect sizes calculated using Cohen’s d (d). In those with PD, Pearson correlational analyses were completed for the relationship between cognitive and motor functioning and the frequency of stuttered disfluencies, disease duration and levodopa usage. Mean differences in the frequency of stuttered disfluencies

across groups and speech tasks were assessed using a two by three mixed-design ANOVA with a log transformation and using bootstrapping procedures with the between-subject factor group (control vs. PD) and within-subject factor task (conversation vs. picture description vs. reading), followed by Eta Squared (η^2) calculations. Bootstrapping of ANOVA model followed procedures described in Spsychala et al.⁴⁹ Post-hoc analyses following recommendations in Field et al.⁵⁰ were completed to investigate task dependent difference, irrespective of the interaction term, using Wilcoxon rank-sum tests and Cohen’s d with Bonferroni corrections. The relationship between the frequency of stuttered disfluencies in conversation and motor and cognitive factors in the PD group was also assessed using linear regression analyses, with age and sex as covariates. Fit of the multiple linear regression models with and without cognitive functioning added to the motor functioning model was compared using ANOVA. All analyses were completed in the R statistical environment (version 4.0.3).⁵¹

Results

Cognitive and Motor Functioning

Global Z cognitive scores ranged from −1.87 to 1.25 in the PD group and −0.30 to 1.56 in the control group. These scores were significantly lower in the PD compared to control group (Table 2, $W = 2197$, $P < 0.01$, $d = 1.47$). Forty-one participants with PD were classified as PD-MCI and seven as PDD. UPDRS Part III scores for the PD participants ranged from 8 to 64 (mild-severe, Table 2).⁵² Cognitive and motor functioning were significantly correlated ($r = -0.58$, $P < 0.01$).

Stuttered Disfluencies

Table 2 summarizes the descriptive statistics for both groups’ frequency of stuttered disfluencies across speech tasks. During conversation, the disfluencies ranged from 0.15 to 9.90 %SD (median = 1.83) in the PD group and 0 to 5.51 %SD (median = 0.73) in the control group. Based on conversational data, around five times as many people in the PD group (21%; $n = 20/94$; 6 PD-N, 11 PD-MCI, 3 PDD; 16 Males, 4 Females) met the ≥3% SD criterion for stuttering compared to the control group (4%; $n = 1/25$; one Male). For those with PD, stuttered disfluencies were significantly correlated with time since disease onset ($r = 0.28$, $P < 0.01$), levodopa equivalent dosage ($r = 0.41$, $P < 0.01$), cognition ($r = -0.36$, $P < 0.01$), and motor functioning ($r = 0.34$, $P < 0.01$) (Fig. S1).

ANOVA confirmed that both group ($F_{(1, 100)} = 5.88$, $P = 0.02$, $\eta^2 = 0.06$) and speech task ($F_{(1, 202)} = 119.0$, $P < 0.01$, $\eta^2 = 0.37$) had a significant impact on the occurrence of stuttered disfluencies. The group by task interaction was not significant ($F_{(1, 202)} = 0.16$, $P = 0.68$, $\eta^2 < 0.01$). Post-hoc analyses, with Bonferroni corrections were used to investigate the main effect of group and task. This showed that a group difference in stuttering frequency was present in the conversation and

TABLE 2 Descriptive statistics and group comparisons of behavioral assessment results

Cognitive Functioning, Motor Functioning and Stuttered Disfluencies	PD	Control	P-value (Wilcoxon)	Effect size—Cohen's <i>d</i>
Cognitive status				
Normal/MCI/dementia	52/41/7	25/0/0	–	–
Global Z				
Mean (standard deviation)	–0.20 (0.72)	0.80 (0.51)	<0.01 (2197)	1.47
Range	–1.87–1.25	–0.30–1.56		
UPDRS Part III				
Mean (standard deviation)	37 (13.08)	–	–	–
Range	8–64	–		
Speech Task				
Conversation (%SD)				
<i>n</i>	94	25		
Mean (standard deviation)	2.25 (1.83)	1.18 (1.19)	<0.01 (626)	–0.63
Range	0.15–9.90	0–5.51		
Picture description (%SD)				
<i>n</i>	79	24		
Mean (standard deviation)	1.56 (1.77)	1.19 (1.69)	0.24 (761)	–0.18
Range	0 to 8.00	0 to 6.80		
Reading (%SD)				
<i>n</i>	99	25		
Mean (standard deviation)	0.79 (1.31)	0.20 (0.28)	0.02 (826.5)	–0.49
Range	0 to 8.15	0 to 0.74		

reading tasks, but not during picture description (Table 2 and Fig. 1). Within the PD group a higher frequency of stuttered disfluencies was present in conversation compared to reading ($W = 8399$, $P < 0.01$, $d = 0.81$) and compared to picture description ($W = 6355$, $P < 0.01$, $d = 0.22$); and in picture description compared to reading ($W = 7005.1$, $P < 0.01$, $d = 0.52$). In the control group, a higher frequency of stuttered disfluencies occurred in conversation compared to reading ($W = 442$, $P < 0.01$, $d = 1.11$); picture description compared to reading ($W = 393.5$, $P < 0.01$, $d = 0.83$); but not in conversation compared to picture description ($W = 305$, $P = 0.38$, $d = -0.02$).

Association Between Stuttered Disfluencies, Cognitive and Motor Functioning

Analysis using multiple regression accounting for age and sex in the PD group showed that cognitive ($F_{(3, 90)} = 5.77$, $P < 0.01$, residual standard error (RSE) = 1.70, adjusted $R^2 = 0.13$) and

motor functioning ($F_{(3, 84)} = 4.96$, $P < 0.01$, RSE = 1.74, adjusted $R^2 = 0.12$) were both significant predictors of the frequency of stuttered disfluencies in conversation (Figs. 2 and 3). The cognitive model was still significant when participants with a classification of dementia were removed ($F_{(3, 84)} = 4.28$, $P < 0.01$, RSE = 1.64, adjusted $R^2 = 0.10$). The combined model ($F_{(4, 83)} = 5.21$, $P < 0.01$, RSE = 1.70, adjusted $R^2 = 0.16$) with cognitive ($P = 0.03$) and motor functioning ($P = 0.3$) significantly improved the prediction of stuttered disfluencies in conversation ($F_{(1, 83)} = 5.21$, $P = 0.03$) compared to the motor-only model.

Discussion

In a sample of 100 PD and 25 control participants, we showed that PD was associated with a significantly increased occurrence of acquired stuttered disfluencies. In addition, increased presence of stuttered disfluencies was associated with poorer motor and cognitive functioning.

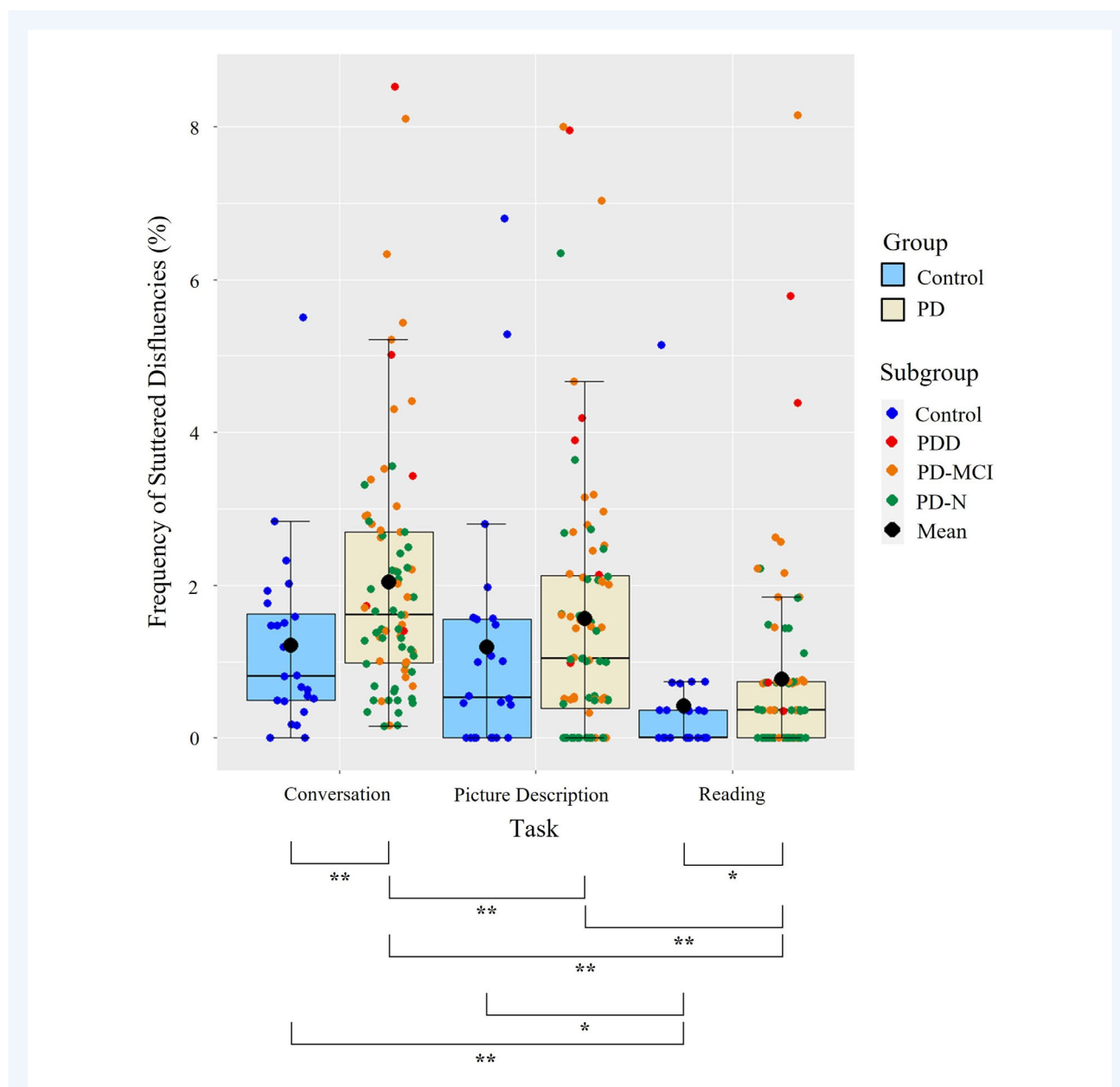


Figure 1. Frequency of stuttered disfluencies in each task by group, divided into subgroups based on cognitive status; colored boxes represent lower quartile, median and upper quartile; PDD, PD dementia; PD-MCI, PD mild cognitive impairment; PD-N, PD normal cognition; **P* < 0.05, ***P* < 0.01.

More specifically, between group comparisons showed that stuttered disfluencies occurred almost twice as often in the group with PD compared to healthy controls in conversation. All types of stuttered disfluencies (prolongations, blocks, part-word and monosyllabic word repetitions) were present in these speech samples. Significant between group differences were also identified in the reading task, with approximately four times as many stuttered disfluencies in the PD group compared to the control group. While Goberman et al did not report conversation data, their reading data similarly showed more than five times as many disfluencies in their 32 participants with PD ($3.3\% \pm 3.4\%$) compared to 10 healthy controls ($0.5\% \pm 0.5\%$).²⁵ However, their

stuttering frequency results during reading were much higher than those in the current study ($SD_{PD} = 0.79\% \pm 1.31\%$; $SD_{Control} = 0.2\% \pm 0.28\%$). This difference does not seem attributable to divergences in disease severity as the both studies included participants across the full severity range.²⁵ The discrepancy may be attributable—in part—to methodological differences, as Goberman et al²⁵ calculated the %SD per 100 words, leading to a higher frequency of disfluencies compared to syllable-based calculations that underpin the 3% criterion.⁴⁷

Next, we determined how many individuals with PD would meet the 3% SD criterion for acquired (new onset) stuttering. Of the 94 PD participants with conversation samples, 20 (21%) were

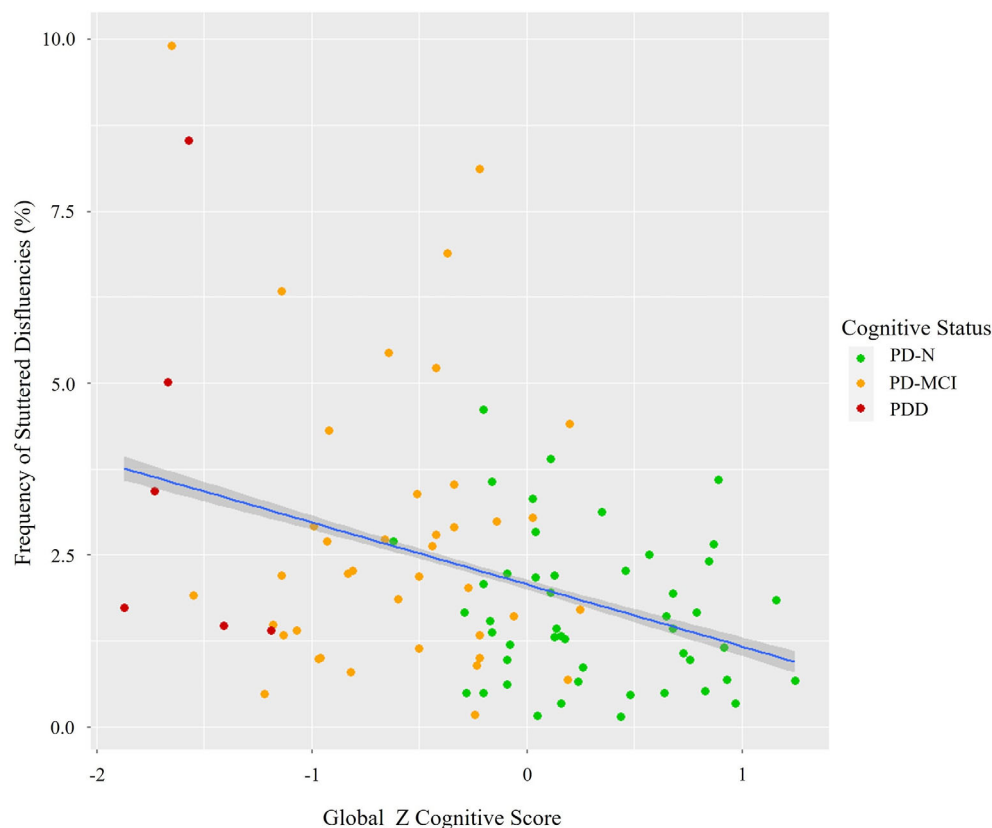
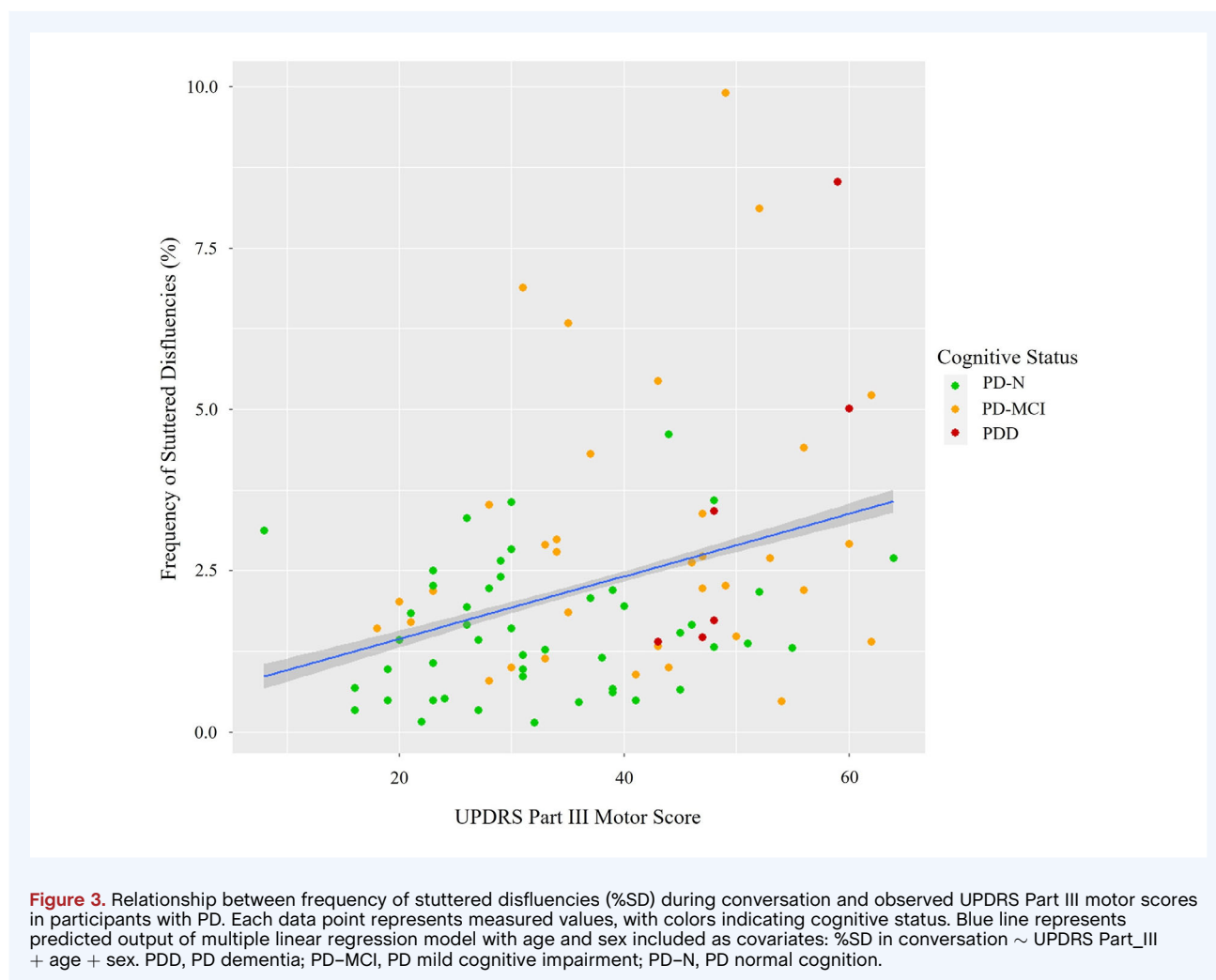


Figure 2. Relationship between frequency of stuttered disfluencies (%SD) during conversation and observed global Z cognitive scores in participants with PD. Each data point represents measured values, with colors indicating cognitive status. Blue line represents predicted output of multiple linear regression model with age and sex included as covariates: %SD in conversation \sim Global_Z + age + sex. PDD, PD dementia; PD-MCI, PD mild cognitive impairment; PD-N, PD normal cognition.

diagnosed with stuttering. Based on the reading task only, 5 PD participants (5%) in our study met the 3% SD criterion. Again, this finding is much lower than the 53% of participants identified with stuttering in Goberman et al's study.²⁵ In addition to the two prospective studies by Goberman et al²⁵ and Juste et al,²⁶ two retrospective database studies reported on the occurrence of disfluencies in PD. The first study identified repetitive speech disorders in 58% of 113 PD participants.³ However, the number of individuals who stuttered was likely much lower as they also captured non-stuttering disfluencies (eg, palilalia, sentence repetitions) in their rating scale, but excluded blocks.³ In contrast, the second retrospective study showed that 4% of 280 PD participants self-reported new-onset stuttering, a finding five times lower than the 21% identified in current study.² Taken together, the wide range from 4 to 53% in identification of stuttering across studies shows the importance of detailed speech analysis using established methods in stuttering research. Despite their differences, these studies provide clear evidence that a subgroup of people with PD will present clinically with acquired neurogenic stuttering. Stuttering is therefore important to consider in the clinical management of communication problems of people with PD.

Between-task comparisons showed that conversation resulted in the highest frequency of stuttered disfluencies, followed by picture description, and then reading across PD and controls. These results are consistent with Juste et al's, who identified a higher frequency of stuttered disfluencies during monologue (ie, speaking continuously on a topic, $3.9\% \pm 4.2\%$ SD) compared to reading ($1.8\% \pm 2.0\%$ SD) in their PD group ($n = 20$).²⁶ The current study provides additional information about conversational speech, which is more representative of natural speaking situations. The task-dependency of acquired stuttering disfluencies is an important consideration for diagnostic purposes, and shows that conversations are the most sensitive task for the identification of stuttered disfluencies in PD. Conversation identified the most participants ($n = 20$) presenting with more than 3% SD. Picture description identified 10 participants and reading identified five participants with more than 3% SD. Three participants identified with more than 3% SD in the monologue task had %SD of 2.5, 2.19 and 1.4 respectively during conversation. It is known that certain speech tasks can have fluency-inducing effects. This mechanism forms the basis for a number of stuttering treatments.⁵³ As our findings indicate task dependent differences in the population of people with PD who stutter,



further investigation of the effect speech tasks such as rhythmic speech, singing and automated speech (eg, naming days of the week) may provide important insights to guide treatment directions in this population.

In those with PD the frequency of stuttered disfluencies during conversation was significantly associated with the disease progression variables, time since symptom onset, and levodopa equivalent dosage. This finding differs from previous findings of Im and colleagues who did not find a significant correlation between duration of PD, duration of levodopa use, or levodopa equivalent dosage.²⁹ However, this may be a consequence of methodological differences. In this present study nearly all participants were seen during the on state of typical dopaminergic medication usage (ranging from 150 to 2900 mg LED). In contrast, in Im's study on state assessment occurred after all PD participants received a controlled dose of 300 mg of levodopa. In addition, Im and colleagues included interjections, phrase repetitions and revisions in their disfluency calculations, which are excluded in our stuttering analyses.²⁹ The current findings are in line with those from Tykalová et al based on cumulative dosages of dopaminergic medication.⁴ They indicate that as the

neurodegeneration progresses over time and a higher dosage of dopaminergic medication is needed to manage symptoms, the frequency of stuttered disfluencies also increases. However, we cannot infer that this is a direct causal relationship as previous research has shown both increases and decreases in stuttered disfluencies in PD cases following adjustments in dopaminergic medications,²⁹ and with deep brain stimulation.^{54,55} Additional symptom severity variables also contribute to this correlation.⁴ Of those, we set out to investigate the influence of motor and cognitive functioning.

A higher frequency of stuttered disfluencies was associated with poorer performance on both cognitive and clinical motor assessment. A combined cognitive and motor model significantly improved prediction of the frequency of stuttered disfluencies compared to accounting for motor functioning alone, with cognition carrying most of the predictive value. This challenges the suggestion that the occurrence of stuttered disfluencies is purely motoric, due to similarities with other motor characteristics in PD.^{25,26} Instead, our findings show that both motor and cognitive factors can have a significant impact on stuttering. This new observation is in line with recent studies that have identified both

motor and cognitive function as contributors to other communication changes in PD (eg, voice, language).^{31,32,34} As suggested previously, difficulty initiating speech motor commands could indeed be secondary to basal ganglia dysfunction, associated with the fluctuations in dopamine levels that occur in PD,^{25,26} however a more complex relationship is also possible. As neurodegeneration expands with disease progression, additional and worsening symptoms, including cognitive decline, appear to further impact communicative functions.²² Ultimately, these findings emphasize that stuttered disfluencies in PD should not be considered in isolation or to be solely related to motor functioning. Instead stuttering should be considered within the broader context of each person's specific presentation of symptoms.

Having identified the presence of stuttered disfluencies in conversational speech of people with PD, next steps would be to investigate the impact of these stuttered disfluencies on overall communication, life participation, and quality of life of people with PD. This is particularly important given the overlap in the frequency of stuttered disfluencies and cognitive functioning between the PD and control group in this study. A further step would be to investigate the influence of specific aspects of motor and cognitive functioning such as dyskinesia and language on the frequency of stuttering. In addition to mapping such relationships in large prospective studies, use of brain imaging technology to investigate the association with behavioral and neural changes may also help to elucidate the mechanisms underlying the development of stuttered disfluencies in this population.^{11,12}

Conclusion

This study found that PD was associated with a significant increase in stuttered disfluencies, with one fifth of PD participants meeting the diagnostic criterion for acquired neurogenic stuttering. The speech task had a significant impact on the occurrence of stuttered disfluencies, with most disfluencies occurring during conversation. An increase in stuttered disfluencies was significantly associated with reduced motor functioning and poorer cognitive scores. This challenges previous suggestions that the development of stuttered disfluencies in Parkinson's disease has a purely motoric basis. Together, these results highlight the importance of assessing stuttered disfluencies when monitoring communication changes in people with PD and suggest that appropriate follow-up and treatment protocols should be put in place.

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T.J.A.: 2C, 3B

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Supporting Information

Supporting information may be found in the online version of this article.

Table S1. Tests used for assessment of each cognitive domain in the neuropsychological battery.

Figure S1. Relationship between frequency of stuttered disfluencies (%SD) during conversation in participants with PD and (A) time since disease onset (years), (B) levodopa equivalent dose (mg), (C) global Z cognitive score, and (D) UPDRS Part III motor scores. Blue line represents correlation between the two variables in each graph. PDD, PD dementia; PD-MCI, PD mild cognitive impairment; PD-N, PD normal cognition.