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Research Article

The Impact of Parkinson's Disease on Breath Pauses and Their Relationship to Speech Impairment: A Longitudinal Study

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Purpose: The purposes of this longitudinal study were to (a) examine the impact of Parkinson's disease (PD) progression on breath pause patterns and speech and linguistic errors and (b) determine the extent to which breath pauses and speech and linguistic errors contribute to speech impairment.

Method: Eight individuals with PD and eight age- and sex-matched control participants produced a reading passage on two occasions (Time 1 and Time 2) 3 years and 7 months apart on average. Two speech-language pathologists rated the severity of speech impairment for all participants at each time. Dependent variables included the location of each breath pause relative to syntax and punctuation as well as the number of disfluencies and mazes.

Results: At Time 1, there were no significant differences between the groups regarding breath pause patterns. At Time 2, individuals with PD produced significantly fewer breath pauses at major syntactic boundaries and periods as well as significantly more breath pauses at locations with no punctuation than control participants. Individuals with PD produced a significantly greater number of disfluencies than control participants at both time points. There were no significant differences between the groups in the number of mazes produced at either time point. Together, the number of mazes and the percentage of breath pauses at locations with no punctuation explained 50% of the variance associated with the ratings of severity of speech impairment.

Conclusion: These results highlight the importance of targeting both respiratory physiological and cognitive–linguistic systems in order to improve speech production in individuals with PD.

Inappropriate silences or pauses are a hallmark characteristic of hypokinetic dysarthria, the dysarthria most often associated with Parkinson's disease (PD; Darley et al., 1969). Since the seminal work of Darley et al. (1969), researchers have sought to quantify the appropriateness of pauses in hypokinetic dysarthria and determine how pauses at unexpected locations contribute to the severity of speech impairment. One approach to this issue has been to classify breath pauses based on syntactic appropriateness. In this approach, pauses in which the speaker takes a breath (hereafter referred to as "breath pauses") are categorized based on the syntax of a passage (Bunton, 2005; Hammen & Yorkston, 1994; Huber et al., 2012; Wang et al., 2005). Specifically, breaths that occur at major or minor syntactic boundaries are considered appropriate; other breath pauses are deemed inappropriate. Since punctuation in written language corresponds closely with syntax (i.e., periods often represent major syntactic boundaries and commas often represent minor syntactic boundaries), periods and commas can also be used to determine the appropriateness of breath pauses when reading (Conrad et al., 1983; Huber et al., 2012).

This approach is a promising objective characterization of inappropriate silences or pauses due to the role that breath pauses play in successful communication. The coordination between breath pauses, syntax, and punctuation allows listeners to use breath pauses to parse running speech into meaningful units. Previous studies have demonstrated that typical speakers pause at syntactically appropriate locations and at locations marked by punctuation and rarely pause at locations that are unrelated to syntax (Grosjean & Collins, 1979; Huber et al., 2012; Price et al., 1991; Wang et al., 2005; Winkworth et al., 1994). Some researchers have suggested that pauses are more important in understanding

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syntactically ambiguous sentences than pitch contours (Price et al., 1991; Shah et al., 2006).

The relationship between breath pauses, syntax, and punctuation requires the careful coordination of both the respiratory physiological and cognitive–linguistic systems (Huber et al., 2012). The purpose of the respiratory subsystem during speech production is to provide steady, driving pressure throughout an utterance regardless of its length. To account for the variability in utterance length (the number of words or syllables that are said on one breath), healthy adults prepare for longer utterances by initiating and terminating speech at higher lung volumes (Bunton, 2005; Huber, 2008; Huber & Darling, 2011; Sperry & Klich, 1992; Winkworth et al., 1994). This preplanning ensures that breath pauses are not taken at locations that are unrelated to a syntactic boundary or punctuation due solely to the physiological need to breathe.

Even as the respiratory physiological system changes with age (i.e., reduced elastic recoil of the lungs, reduced chest wall compliance, and reduced respiratory muscle strength) and as initiating speech at higher lung volumes becomes more difficult (Huber, 2008), older adults maintain this preplanning behavior and compensate for respiratory physiological changes by reducing utterance length (Huber & Darling, 2011). As a result, older adults increase the number of breath pauses at minor syntactic boundaries and commas but do not increase breaths at locations unrelated to a syntactic boundary, as utterance length decreases with age (Huber et al., 2012). Thus, healthy speakers maintain the relationship between breath pauses and syntax throughout the life span.

Individuals with PD demonstrate both age- and disease-related changes to the respiratory physiological system. When compared to age- and sex-matched older adults, individuals with PD demonstrate changes in pulmonary function as evidenced by reduced forced vital capacity and forced expiratory volume in 1 s (De Pandis et al., 2002), increased chest wall rigidity (Sabaté et al., 1996; Solomon & Hixon, 1993), and reduced respiratory muscle strength and coordination (De Bruin et al., 1993; Haas et al., 2004; Pitts et al., 2008; Weiner et al., 2002). These physiological impairments result in abnormal speech breathing patterns, such as initiating and terminating speech at higher- or lower-than-normal lung volumes, that lead to an overreliance on active respiratory muscle forces and increased work of speech breathing (Bunton, 2005; Huber & Darling, 2011; Huber & Darling-White, 2017; Solomon & Hixon, 1993). Individuals with PD also demonstrate weak relationships between lung volume initiation and utterance length, suggesting that something about the disease process is interfering with the ability to coordinate the respiratory physiological and cognitive–linguistic systems to plan appropriate respiratory support for an utterance (Huber & Darling, 2011). A lack of appropriate respiratory support in combination with effortful, fatiguing speech production may lead to individuals with PD pausing out of the physiological need to breathe regardless of syntactic location (Huber et al., 2012). Thus, it is no surprise that the relationship between breath pauses and syntax is disrupted in individuals with PD (Bunton, 2005; Hammen & Yorkston, 1994; Huber et al., 2012).

Data regarding the impact of PD on breath pauses taken at major and minor syntactic boundaries are mixed with results seemingly mediated by the severity of the speech impairment of the individuals being studied (Huber et al., 2012). An examination of the individual data from these studies indicates that individuals with PD who demonstrate moderate or moderate–severe speech impairment tend to take fewer breath pauses at major syntactic boundaries and more breath pauses at minor syntactic boundaries than healthy older adults (Hammen & Yorkston, 1994). However, individuals with PD who demonstrate mild speech impairment do not differ from healthy older adults in regard to breath pauses at major and minor syntactic boundaries (Huber et al., 2012). Regardless of the severity of the speech impairment, individuals with PD take more breath pauses at locations unrelated to a syntactic boundary than healthy older adults, although this finding appears to be strongest for individuals who demonstrate more moderate speech impairment (Huber et al., 2012).

Our understanding of the impact of PD on the relationship between breath pauses and punctuation is still highly preliminary. Huber et al. (2012), the only study to investigate this relationship in individuals with PD and healthy older adults, found no significant group differences. However, mean data indicate that individuals with PD took more breath pauses at locations with no punctuation than healthy older adults. However, the majority of the individuals with PD in this study had relatively mild speech impairments, and it is possible that abnormal breath pausing would be present in individuals with more severe speech impairments.

Despite the apparent link between the severity of the speech impairment and the coordination of breath pauses with syntax and punctuation, there have been no empirical studies directly examining this relationship. One possible way to investigate this relationship is through the lens of disease progression given that the severity of the speech impairment worsens with disease progression in individuals with PD (Huber & Darling-White, 2017; Skodda et al., 2013). When examining disease progression in individuals with PD, longitudinal research designs are well suited to differentiate the role of disease from the role of aging. However, longitudinal studies regarding the impact of PD on speech production are lacking. This is likely due to the challenges this type of design imposes. In order to differentiate between the impact of aging and the impact of disease, longitudinal studies must include both healthy older adults and individuals with PD. Participant attrition in any longitudinal study is common due to reasons such as relocation, but given the typical age of both of these groups (> 65) and the fact that PD is degenerative, participant attrition due to death and/or illness is a significant hindrance to this type of work. It can also be difficult to draw strong conclusions about the impact of disease when variables related to disease management (e.g., medication changes, deep brain stimulation, participation in speech therapy) are uncontrollable over the course of several years. Despite these challenges, longitudinal studies are vital to the development of evidence-based interventions for each stage of the disease. Longitudinal studies may also help identify markers of disease progression in the speech production system. This is particularly important in a disorder where declines in speech and language production do not necessarily correlate with declines in motor function (Ash et al., 2017; Skodda et al., 2011, 2013, 2009).

The current study applied the breath pause classification scheme found in the work of Huber et al. (2012) to the longitudinal data set presented in the work of Huber and Darling-White (2017) in order to extend our understanding of the impact of PD on the coordination of breath pauses with syntax and punctuation. Eight individuals with PD and eight age- and sex-matched control participants produced two trials of a reading passage on two separate occasions 3 years and 7 months apart on average (hereafter referred to as "Time 1" and "Time 2"). The severity of the speech impairment significantly increased over time for the individuals with PD, but not control participants, and the individuals with PD represented a wide range of speech impairments. Utterance length did not significantly change across time or differ between the groups. Thus, any changes in breath pause patterns found in this study are likely the result of disease progression and not differences in utterance length. Additionally, speech breathing patterns were significantly different over time and between the groups. With disease progression, individuals with PD demonstrated significant decreases in lung volume initiation and termination leading to more effortful speech production due to increased reliance on expiratory muscle forces. Since respiratory physiological factors contribute to the coordination of breath pauses with syntax and punctuation in individuals with PD, breath pause patterns may change with disease progression as a result of changes in respiratory physiology.

The classification scheme originally published in the work of Huber et al. (2012) includes the identification of speech errors such as disfluencies (any single-sound or single-word repetition) and linguistic errors such as mazes (multiple word repetitions, additions to the reading passage, and other deviations from the passage content), in addition to the classification of breath pauses by syntax and punctuation. As compared to age- and sex-matched older adults, individuals with PD did not produce significantly different numbers of disfluencies but did produce significantly more mazes (Huber et al., 2012). Since reading tasks require very little language formulation, deviations from the text in the form of linguistic errors may be indicative of language processing and/or formulation impairments.

Although very little is known about the impact of disease progression on speech or linguistic errors, evidence from longitudinal studies presents a different picture than the cross-sectional study by Huber et al. (2012). Disfluencies appear to increase over time in all individuals with PD (Tykalová et al., 2015), but only individuals with PD and evidence of dementia demonstrate significant declines in fluency, grammatical structure, and the level of informativeness during a picture description task over time (Ash et al., 2017). Examining the impact of disease progression on speech and linguistic errors will allow for a more complete understanding of the factors that potentially contribute to the severity of the speech impairment in individuals with PD.

The primary purpose of this longitudinal study was to examine the impact of disease progression on the coordination of breath pauses with syntax and punctuation as well as on the production of speech and linguistic errors. These data will be vital to the development of evidence-based interventions for each stage of the disease. The secondary purpose was to determine the extent to which breath pauses and speech and linguistic errors contribute to ratings of speech impairment. While the variables that contribute to ratings of speech impairment are highly complex, it is important to begin the process of untangling these relationships. By doing so, we hope to identify specific targets for intervention. If speech-language pathologists (SLPs) can focus on the most important aspects of speech impairment in individuals with PD, it is possible that interventions will be more efficient and effective.

We addressed the following research questions:

1. Does the coordination of breath pauses with syntax and punctuation change between the groups (individuals with PD and control participants) over time? On the basis of the findings of Huber et al. (2012), we hypothesized that, at Time 1, individuals with PD would produce more breath pauses at locations unrelated to a syntactic boundary than control participants. We expected the differences between the groups to be greater at Time 2. In addition to more pauses at locations unrelated to syntax, we hypothesized that individuals with PD would produce fewer breath pauses at major syntactic boundaries and periods than control participants at Time 2.
2. Does the production of speech and linguistic errors change between the groups (individuals with PD and control participants) over time? We hypothesized that individuals with PD would produce a greater number of disfluencies and mazes than control participants at both Time 1 and Time 2.
3. Do the location of breath pauses and speech and linguistic errors contribute to ratings of speech impairment? We hypothesized that more breath pauses unrelated to a syntactic boundary or punctuation and greater numbers of speech and linguistic errors would be correlated with higher ratings of speech impairment.

Method

Research Design

This longitudinal study included acoustic and kinematic data (described below) from two separate data collection sessions. The average period between data collection sessions was 3 years and 7 months (*SD* = 6.5 months). Data collection sessions occurred in the Speech Physiology Laboratory at Purdue University. This study was approved by the Purdue University Institutional Review Board. Data from Time 1 were collected as part of a larger cross-sectional study examining the impact of different types of cues to increase loudness on speech production patterns and were presented in the work of Huber et al. (2012). However, Participant F07PD was not included in that data set. At Time 1, Participant F07PD had not been diagnosed with PD and was recruited into the control participant group. Her data were excluded from publication because she was a significant outlier. During the recruitment process for Time 2, Participant F07PD disclosed that she had been diagnosed with PD a few months prior. The pathophysiological changes characteristic of PD begin prior to clinically noticeable symptoms (Bernheimer et al., 1973). This results in a period of time (often many years) in which individuals may exhibit subtle behavioral changes that do not reach the level of clinical significance. In a single case study, an individual with PD was found to have significant changes in fundamental frequency variability for the 5-year period prior to diagnosis (Harel et al., 2004). Since Participant F07PD's Time 1 data fit the profile of an individual with PD and she was given the diagnosis prior to Time 2, we included her in the longitudinal data set presented in the work of Huber and Darling-White (2017) and in the current study. Data from Time 2, with the exception of the speech impairment ratings, have not been previously published. Speech impairment ratings were initially presented in the work of Huber and Darling-White.

Participants

Eight individuals diagnosed with idiopathic PD (four men, four women) and eight age- and sex-matched older adults (i.e., control participants) were involved in this longitudinal study. The same individuals were examined at both time points. Individuals with PD were between ages 68;9 (years;months) and 80;0 (*M* = 72;9, *SD* = 4;1) at Time 1 and between ages 72;2 and 85;2 (*M* = 76;3, *SD* = 4;0) at Time 2. Control participants were between ages 65;8 and 82;0 (*M* = 72;10, *SD* = 4;7) at Time 1 and between ages 69;7 and 85;6 (*M* = 76;7, *SD* = 4;6) at Time 2. There were no significant differences in age between the groups at either time point (Time 1: $t = -0.06$, $p = .955$; Time 2: $t = 0.16$, $p = .879$).

The inclusion criteria for Time 1 were as follows: (a) no history of respiratory problems or neurological disease (except PD); (b) no history of head, neck, or chest cancer or surgery; (c) no formal training in singing or speaking; (d) nonsmoking for at least the past 5 years; (e) ambulatory and living independently in the community; (f) adequate cognition as measured by a score of 24 or above on the Mini-Mental State Examination (Folstein et al., 1975); and (g) free of infections, colds, and allergies on the day of testing. Additionally, the age- and sex-matched older adults were required to demonstrate normal speech, language, and voice as determined by participant self-report and by the subjective judgment of the second author, a certified SLP, and to pass a bilateral hearing screening at 40 dB HL for 500, 1000, and 2000 Hz (Ventry & Weinstein, 1983). Although hearing status was not an inclusion criterion for individuals with PD at Time 1, all participants with PD passed the hearing screening except for Participant M04PD who did not pass at 40 dB HL for 2000 Hz in the right ear. Only one individual with PD (Participant M09PD) reported receiving speech therapy services. He participated in weekly group speech therapy sessions for 2 years prior to Time 1. No other participants (control participants included) had a history of speech therapy within 20 years of Time 1.

For the Time 2 data collection session, only those pairs (an individual with PD and their age- and sex-matched older adult) for which both members of the pair from Time 1 could participate in Time 2 were included in the current study. At Time 2, all participants were still nonsmoking and living independently in the community. A measure of overall severity of motor involvement in individuals with PD, such as the Hoehn and Yahr scale, was not available. However, since all participants were living independently in the community throughout the study, individuals with PD were within Hoehn and Yahr stages I–IV. It is important to note that changes in speech production in individuals with PD are often independent of changes in gross motor function (Ash et al., 2017; Skodda et al., 2011, 2009). Participants were free of infections, colds, and allergies on the day of testing. Control participants and the majority of participants with PD denied any respiratory or neurological issues (except PD) and recent head, neck, or chest surgeries. Between Time 1 and Time 2, Participant M04PD underwent deep brain stimulation. No other participants with PD had deep brain stimulation. Participant F01PD had a possible transient ischemic attack 1 year prior to Time 2 data collection, but this was not definitively diagnosed. Cognition was assessed using the Cognitive Linguistic Quick Test (CLQT; Helm-Estabrooks, 2001) at Time 2. All control participants and the majority of participants with PD continued to demonstrate typical cognition based on the CLQT composite score. Two participants with PD demonstrated cognitive decline from Time 1 to Time 2. Participant M09PD had a moderate cognitive deficit, and Participant M10PD had a mild cognitive deficit based on the CLQT composite score at Time 2. Hearing status remained the same for all participants between Time 1 and Time 2, except for Participant M07OC who did not pass at 40 dB HL for 1000 and 2000 Hz in the right ear. Five individuals with PD were receiving or had received some speech-language therapy between Time 1 and Time 2 for reasons such as word-finding problems (M10PD), decreased loudness (F01PD, F02PD, M04PD), decreased clarity of speech (M09PD), and abnormal speech rate (F01PD). Age- and sex-matched older adults continued to demonstrate normal speech, language, and voice, and no one reported speech or language therapy between Time 1 and Time 2. Demographic information, including age, years since diagnosis, medications, and CLQT composite scores, is presented in Table 1.

Ratings of Speech Impairment

Two certified SLPs, who were not affiliated with the study, completed ratings of speech impairment for both groups (individuals with PD and control participants; see Table 1). The SLPs were experienced in the assessment and treatment of motor speech disorders but were not familiar with the participants in this study. Using a visual analog, with one end labeled "normal" and the other end labeled "very severe," the SLPs rated the severity of the speech impairment from the middle 30 s of a spontaneous speech task. In the spontaneous speech task, participants were asked to talk about a topic of their choice (e.g., a recent vacation, family, pets) for approximately 2 min. Since the study used experienced raters who have worked with patients with neurological diseases across a range of severities, perceptual anchors were not provided. Individual audio files were created, and amplitude was normalized to 70 dB using Praat (Boersma & Weenink, 2008). The SLPs listened to the speech samples via headphones at a self-selected output level. They were asked to use one output level for the entire task. The SLPs were blinded to disease state (PD vs. control) and data collection point (Time 1, Time 2). The speech samples were randomized within blocks of speaker sex (females presented first and then males). The SLPs were allowed to listen to each speech sample one time. Ratings were measured in millimeters from the "normal" end of the visual analog scale to the line marked by the SLP and then converted into a percentage by dividing the rating in millimeters by the total millimeters of the visual analog scale and multiplying by 100. A higher number indicates a more severe speech impairment. The percentages were averaged for each participant and each time point. If the difference between the SLP ratings was greater than 20%, the second author, a certified SLP, rated the sample. This occurred for seven of the 43 samples. This third rating was then averaged with the original rating that was closest.

Equipment

Respiratory kinematic data were collected using the Respitrace (Ambulatory Monitoring, Inc.). Two elastic bands, one placed around the rib cage just under the axilla and one placed below the last rib at the level of the participant's umbilicus, were used to transduce the signals from the rib cage and the abdomen, respectively. Acoustic data were collected using a high-quality condenser microphone (Time 1: Brüel & Kjær Type 4936; Time 2: Countryman E6 Model E610P5L2). The microphone signal was digitized and time-locked to the respiratory signal.

Procedures and Speech Stimuli

Each participant read "The Papa Passage" two times in each data collection session at a comfortable loudness level and pitch (Sapienza & Stathopoulos, 1995). The passage is 68 words and 12 sentences long. The reading passage was presented on a computer screen placed at eye level approximately 2 ft from each participant. While there were a variety of different tasks collected during each data collection session, the reading passage was collected as one of the first speech tasks at each time point.

Measurements

The location of each breath pause was determined by a sharp upward deflection in the sum signal from the Respitrace (Huber et al., 2012). The sum signal was computed by summing the calibrated rib cage and abdominal signals using customized MATLAB (MathWorks) programs (Huber, 2007, 2008; Huber et al., 2005; Huber & Darling, 2011). In cases where the identification of a breath pause was difficult, the microphone signal was used to provide additional cues. If there was a silent period in the microphone signal during an upward deflection of the sum signal, a breath pause was recorded. To be consistent with the work of Huber et al. (2012), the location of each breath pause was analyzed based on the punctuation and syntax of the reading passage. Breath pauses in each category were expressed as a percentage of the total number of breath pauses taken during the reading passage. These measurements were made for each participant and each trial from each data collection session.

Syntactic Analysis

Breath pauses were grouped into three distinct categories: breath pauses taken at major syntactic boundaries, breath pauses taken at minor syntactic boundaries, and breath pauses taken at locations unrelated to a syntactic boundary (Huber et al., 2012). Breath pauses at major syntactic boundaries were defined as any breath pause taken after an independent clause. Breath pauses at minor syntactic boundaries were defined as any breath pause taken after a dependent clause or before a prepositional phrase. Breath pauses at locations unrelated to a syntactic boundary were defined as any breath pause that did not fall into the two previous categories (e.g., in the middle of a prepositional phrase, in the middle of a word, after a pronominal subject). A copy of the reading passage, with syntactic boundaries marked, is provided in the work of Huber et al. (2012).

Punctuation Analysis

Breath pauses were grouped into three distinct categories: breath pauses taken at periods, breath pauses taken at commas, and breath pauses taken at locations with no punctuation.

Error Analysis

Two researchers independently listened to each reading passage and orthographically transcribed deviations

Table 1. Participant demographic information.

Participant	Age at Time 1 (years;months)	Age at Time 2 (years;months)	Years since diagnosis (Time 1)	Medications at Time 1	Medications at Time 2	CLQT at Time 2	Speech impairment at Time 1 (%)	Speech impairment at Time 2 (%)
F02OC	74;5	78;6		Procardia, Avapro, Amaryl, Glucophage	Procardia, Avapro, Amaryl, Glucophage	WNL	1.3	4.7
F05OC	73;1	77;1		Lipitor	Lipitor, Diazepam	WNL	3.5	7.1
F07OC	65;8	69;7		None	Estrace	WNL	1.0	1.0
F13OC	73;5	76;9		Atenolol, Norvasc, Lipitor	Atenolol, Norvasc, Lipitor, Fosamax	WNL	4.5	2.5
M06OC	70;6	74;1		None	None	WNL	0.6	2.0
M07OC	70;6	74;0		None	Eye drops for glaucoma	WNL	3.5	7.5
M09OC	82;0	85;6		Lipitor	Lipitor	WNL	7.5	0.3
M11OC	73;5	77;3		Aspirin	None	WNL	0.3	3.0
F01PD	72;5	76;3	0.75	Mirapex, Prozac	Sinemet, Mirapex, Prozac, Buspirone, Aspirin	WNL	8.5	50.5
F02PD	69;9	72;2	9.0	Sinemet, Eldepryl, Clinoril, Zoloft, Welbutrin, Maxide, Tylenol	Sinemet, Eldepryl, Clinoril, Zoloft, Lipitor, Avapro, Inderal, Detrol, Aspirin	WNL	12.0	43.0
F04PD	74;3	76;11	5.0	Sinemet, Eldepryl, Bromocriptine	Sinemet, Bromocriptine, Zelapar	WNL	2.8	11.4
F07PD	72;2	75;11	−3.0[a]	None	Prilosec	WNL	3.5	10.0
M04PD	68;9	73;5	3.5	Stalevo, Permax	Sinemet, Sinemet CR, Requip, Flomax	WNL	37.5	82.0
M09PD	72;8	76;8	9.0	Sinemet, Lipitor, Prozac, Metoprolol	Sinemet, Aricept, Lipitor, Metoprolol, Tylenol, Ibuprofen	Moderate	35.5	73.0
M10PD	70;0	73;7	4.5	Sinemet	Sinemet, Aricept, Mirtazapine, Donepezil	Mild	3.0	8.9
M11PD	82;0	85;2	3.75	Amantadine, Sinemet, Carvedilol, Flomax	Amantadine, Sinemet, Lodosyn, Carvedilol, Provigil, Clonazepam, Fosamax, Flomax	WNL	43.0	35.5

Note. Higher numbers indicate more severe speech ratings for speech impairment. n/a means "data not applicable." CLQT = Cognitive Linguistic Quick Test (Helm-Estabrooks, 2001); F = female; OC = control participant; WNL = within normal limits; M = male; PD = Parkinson's disease.

[a]Diagnosed with PD 3 years after Wave 1.

from the text. When there was a discrepancy between the researchers about what the participant said, the two transcribers came to consensus. Deviations from the text were classified as disfluencies (speech errors) or mazes (linguistic errors). Disfluencies were defined as any single-sound, syllable, or word repetitions. Mazes were defined as multiple-word repetitions, restarted utterances, or any other deviation from the print (Huber & Darling, 2011; Huber et al., 2012). Breath pauses taken during or immediately after a disfluency or maze were not included in the syntactic analysis due to the small numbers of these events. Breath pauses taken during or immediately after a disfluency constituted 0.2% of total breaths at Time 1 and 0.4% of total breaths at Time 2. Breath pauses taken during or immediately after a maze constituted 0.9% of total breaths at Time 1 and 1.1% of total breaths at Time 2.

Reliability

Data from two older adults and two individuals with PD at both time points (4 participants × 2 trials of the reading passage at each time point × 2 time points = 16 total reading passages) were reanalyzed by a second data analyst. For two out of the 16 reading passage samples, one analyst marked one more breath than the other analyst. In total, one analyst marked 121 breath pauses; the other marked 123 breath pauses. These discrepant breath pauses were labeled as occurring at a major syntactic boundary and a minor syntactic boundary. A matched-pairs *t* test was conducted on the number of breaths at major and minor syntactic boundaries to determine if the differences across measurers were significant. The differences were nonsignificant, both *t* tests: $t(7) = 1$, $p = .351$. There were no other differences between the two analysts noted for any of the other dependent variables. Measures were deemed to have acceptable reliability.

Statistical Analysis

All statistical tests were performed using SAS 9.4. Our research questions focused on (a) differences in breath pauses and speech and linguistic errors over time between the groups and (b) the relationship of breath pauses and speech and linguistic errors to ratings of speech impairment.

Nonparametric statistical tests were utilized given the skewed nature of our dependent variables (e.g., clearly nonnormal distributions and high numbers of zeros for some variables). Wilcoxon/Kruskal–Wallis tests were used for the between-group comparisons in each time point. The alpha level for each statistical test was set as $\alpha < .05$.

All data points from Time 1 and Time 2 were included in the following regression analyses. Linear regressions were completed to assess the relationship between ratings of speech impairment and each dependent variable. The alpha level for each statistical test was set as $\alpha < .05$. Dependent variables that were found to have a significant relationship with ratings of speech impairment were entered into a forward and backward stepwise regression model in SAS 9.4. Variables were added one at a time to the model according to the "entry" threshold ($\alpha = .3$). Variables in the model were removed if they did not meet the "stay" threshold ($\alpha = .05$). The stepwise procedure was complete when none of the variables outside the model met the "entry" threshold and every variable in the model met the "stay" threshold.

Results

Means and standard errors for each group and each time point are presented in Table 2. Figure 1 depicts the percentage of breath pauses by syntactic category produced by individuals with PD and control participants at Time 2. Figure 2 depicts the percentage of breath pauses by punctuation category produced by individuals with PD and control participants at Time 2.

Between-Group Differences: Effect of Disease Progression

Syntactic Analysis

There were no significant differences between control participants and individuals with PD for the percentage of breath pauses at major syntactic boundaries at Time 1 ($Z = -1.13$, $p = .259$). At Time 2, individuals with PD took significantly fewer breath pauses at major syntactic boundaries than control participants ($Z = -2.47$, $p = .014$). There were no significant differences between control participants and individuals with PD for the percentage of breath pauses at minor syntactic boundaries (Time 1: $Z = 0.06$, $p = .953$; Time 2: $Z = 1.45$, $p = .141$) or the percentage of breath pauses at locations unrelated to syntax (Time 1: $Z = 1.44$, $p = .150$; Time 2: $Z = 1.83$, $p = .067$) at either time point.

Punctuation Analysis

There were no significant differences between control participants and individuals with PD for the percentage of breath pauses at periods at Time 1 ($Z = -1.11$, $p = .267$). At Time 2, individuals with PD took significantly fewer breath pauses at periods than control participants ($Z = -2.40$, $p = .017$). There were no significant differences between control participants and individuals with PD for the percentage of breath pauses at commas at Time 1 ($Z = -0.24$, $p = .812$) or Time 2 ($Z = 1.04$, $p = .299$). There were no significant differences between control participants and individuals with PD for the percentage of breath pauses at locations with no punctuation at Time 1 ($Z = 1.59$, $p = .111$). At Time 2, individuals with PD took significantly more breath pauses at locations with no punctuation than control participants ($Z = 2.02$, $p = .044$).

Error Analysis

Individuals with PD produced a significantly greater number of disfluencies than control participants at Time 1 ($Z = 2.44$, $p = .015$) and Time 2 ($Z = 2.69$, $p = .007$). There were no significant differences between control participants

Table 2. Means and standard errors (in parentheses) for each group by wave.

Measure	Time 1	Time 2
% of breaths at major syntactic boundaries		
PD	65.80 (3.34)	57.59 (3.78)
Control participants	72.04 (2.32)	69.33 (2.25)
% of breaths at minor syntactic boundaries		
PD	29.73 (3.08)	33.56 (2.89)
Control participants	27.96 (2.32)	28.11 (1.93)
% of breaths at locations unrelated to syntax		
PD	1.85 (1.29)	6.68 (2.51)
Control participants	0 (0)	0.94 (0.65)
% of breaths at periods		
PD	64.08 (3.43)	56.40 (4.11)
Control participants	69.34 (1.99)	68.64 (2.35)
% of breaths at commas		
PD	28.66 (2.81)	30.23 (2.72)
Control participants	29.86 (2.00)	27.69 (1.87)
% of breaths at locations with no punctuation		
PD	4.65 (1.89)	11.20 (3.47)
Control participants	0.79 (0.55)	2.05 (1.00)
Disfluencies		
PD	0.47 (0.19)	0.69 (0.25)
Control participants	0 (0)	0.06 (0.06)
Mazes		
PD	0.73 (0.30)	1.57 (0.66)
Control participants	0.25 (0.11)	0.44 (0.16)

Note. PD = Parkinson's disease.

and individuals with PD for the number of mazes (Time 1: $Z = 1.13$, $p = .259$; Time 2: $Z = 1.07$, $p = .283$).

Relationship Between Speech Impairment and Dependent Variables

Linear Regressions

Syntactic analysis. There was a significant positive relationship between ratings of speech impairment and breath pauses at major syntactic boundaries ($R^2 = .15$, $p = .031$). A lower percentage of breath pauses at major syntactic boundaries was related to ratings of more severe speech impairment. There was no relationship between ratings of speech impairment and breath pauses at minor syntactic boundaries ($R^2 = .008$, $p = .620$). There was a significant positive relationship between ratings of speech impairment and breath pauses at locations unrelated to a syntactic boundary ($R^2 = .26$, $p = .003$). A higher percentage of

Figure 1. Percentage of breath pauses by syntactic category produced by individuals with Parkinson's disease (PD) and control participants at Time 2. MAJ = major syntactic boundaries; MIN = minor syntactic boundaries; UNR = locations unrelated to a syntactic boundary. Error bars indicate standard error. *Significance at $p < .05$.

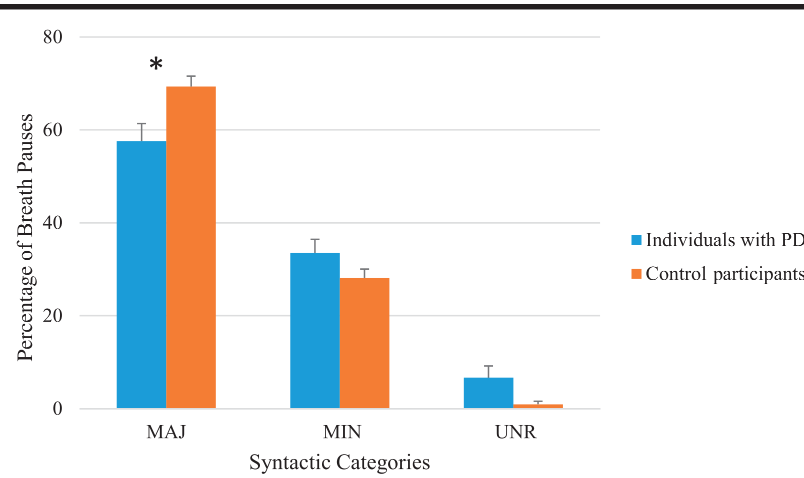
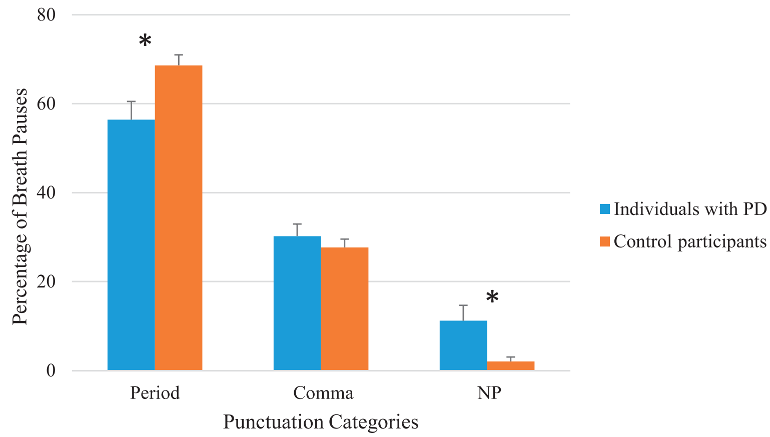


Figure 2. Percentage of breath pauses by punctuation category produced by individuals with Parkinson's disease (PD) and control participants at Time 2. NP = locations with no punctuation. Error bars indicate standard error. *Significance at p < .05.



breath pauses at locations unrelated to a syntactic boundary was related to ratings of more severe speech impairment.

Punctuation analysis. There was a significant positive relationship between ratings of speech impairment and breath pauses at periods (R^2 = .15, p = .029). A lower percentage of breath pauses at periods was related to ratings of more severe speech impairment. There was no relationship between ratings of speech impairment and breath pauses at commas (R^2 = .02, p = .454). There was a significant positive relationship between ratings of speech impairment and breath pauses at locations with no punctuation (R^2 = .32, p < .001). A higher percentage of breath pauses at locations with no punctuation was related to ratings of more severe speech impairment.

Error analysis. There was a significant positive relationship between ratings of speech impairment and disfluencies (R^2 = .36, p < .001). A higher number of disfluencies was related to ratings of more severe speech impairment. There was a significant positive relationship between ratings of speech impairment and mazes (R^2 = .37, p < .001). A higher number of mazes was related to ratings of more severe speech impairment.

Stepwise Regression

The dependent variables that were significantly related to ratings of speech impairment were considered in the stepwise model: breath pauses at major syntactic boundaries, breath pauses at locations unrelated to a syntactic boundary, breaths pauses at periods, breath pauses at locations with no punctuations, mazes, and disfluencies. The first step (R^2 = .37) included mazes (F = 17.28, p < .001). The second step (R^2 = .50) included mazes (F = 10.26, p = .003) and breath pauses at locations with no punctuation (F = 7.49, p = .01). The third step (R^2 = .54) included mazes (F = 13.87, p < .001), breath pauses at locations with no punctuation (F = 7.74, p = .01), and breath pauses at locations unrelated to a syntactic boundary (F = 2.93, p = .10). However, the dependent variable "percentage of breath pauses at locations unrelated to a syntactic boundary" did not meet the "stay" threshold and was subsequently removed from the model. No other variables met the "entry" threshold. Thus, the final model (R^2 = .50) included mazes (F = 10.26, p = .003) and breath pauses at locations with no punctuation (F = 7.49, p = .01).

Discussion

The purposes of this longitudinal study were to examine the impact of disease progression on breath pausing patterns, disfluencies, and mazes and to assess the relationship between these variables and ratings of speech impairment over time. The data clearly demonstrate an impact of disease progression on breath pausing patterns as they relate to both syntax and punctuation. Furthermore, these changes explained some of the variance in ratings of speech impairment across the two time points.

The Effect of Disease Progression

While the differences between the groups were small in Time 1, individuals with PD demonstrated less appropriate breath pausing patterns in Time 2 as compared to control participants. Individuals with PD produced significantly fewer breath pauses at major syntactic boundaries and periods and significantly more breath pauses at locations with no punctuation. Mean data support that individuals with PD produced more breath pauses at locations unrelated to a syntactic boundary than control participants in Time 2 (PD: M = 6.68%; control participants: M = 0.94%), although this did not reach the level of significance (p = .067). On the basis of the findings from the work of Huber and Darling-White (2017), we know that the individuals with PD in the current study initiated and terminated speech at lower lung volumes in Time 2. However, utterance length and lung volume excursion (i.e., the lung volume used for speech production during one breath) did not significantly differ across time. Thus, respiratory physiological changes alone cannot explain the changes in breath pausing patterns observed in this study. Although Huber and Darling-White did not investigate the impact of disease progression on the relationship between lung volume initiation and utterance length, it is possible that individuals with PD had a more difficult time preplanning the respiratory support needed for each utterance as the disease progressed. If individuals with PD were unable to initiate longer utterances at an appropriate lung volume to maintain adequate respiratory support throughout the utterance, they likely needed to breathe at locations unrelated to punctuation or syntax due to physiological need.

As expected, individuals with PD produced a greater number of disfluencies than control participants in both Time 1 and Time 2. Disfluencies were rare for control participants. There were no disfluencies produced by control participants in Time 1. In Time 2, one control participant produced a single disfluency. On the other hand, four out of the eight individuals with PD produced at least one disfluency, for a grand total of seven disfluencies in Time 1. In Time 2, six out of the eight individuals with PD produced at least one disfluency, for a grand total of 11 disfluencies.

The ability to preplan respiratory support for each utterance relies on an intact cognitive–linguistic system and an intact respiratory physiological system. Given the possibility of cognitive–linguistic impairment in individuals with PD (e.g., Alvar et al., 2019; Dick et al., 2018; Huber & Darling, 2011; Illes et al., 1988; Lee, 2017; Murray, 2000; Smith et al., 2018; Troche & Altmann, 2012), we examined the production of mazes. Disease progression did not significantly affect the production of mazes in this study. This was surprising given that previous work from our research team has shown that individuals with PD produce a significantly higher number of mazes than control participants (Huber & Darling, 2011; Huber et al., 2012). The discrepancy between the current results and previous work from our laboratory may be due in part to the number of participants included in each study. Our previous work included 14–15 individuals with PD as opposed to the eight individuals with PD included in this longitudinal study. The individual subject data indicate that many of the individuals with PD did produce a greater number of mazes in both time points as compared to control participants. In Time 1, five out of the eight individuals with PD produced at least one maze, for a grand total of 11 mazes. In Time 2, six out of the eight individuals with PD produced at least one maze, for a grand total of 25 mazes. However, mazes were not as rare as disfluencies for control participants. In Time 1, three out of the eight control participants produced at least one maze, for a grand total of four mazes. In Time 2, six out of the eight control participants produced at least one maze, for a grand total of seven mazes. Thus, it is possible that with more individuals with PD, the effect of maze production would have been stronger. It is also possible that the production of mazes is a normal aspect of our speech production and is not a useful metric when distinguishing healthy aging from disease. In a larger cohort of 25 healthy older adults from the work of Huber et al. (2012), the mean number of mazes produced during the same reading passage was 0.7, a higher average than that for control participants in this study at either time point and the same average as that for individuals with PD at Time 1. In order to interpret the production of mazes by individuals with PD within an appropriate framework, we must first understand the use of mazes by healthy older adults. Thus, future work should focus on examining the use of mazes in large cohorts of healthy older adults.

The utility of mazes as a potential marker of cognitive–linguistic deficits is not clear from these data. All control participants and all but two individuals with PD scored within normal limits on the CLQT at Time 2. Participant M09PD, who had a moderate cognitive deficit based on the CLQT at Time 2, produced more mazes (n = 14) than any other individual with PD at Time 2. However, Participant M10PD, who had a mild cognitive deficit based on the CLQT composite score at Time 2, only produced one maze at Time 2, which was consistent with some of the other individuals with PD and control participants who produced linguistic errors, but did not demonstrate overt cognitive–linguistic deficits. Future studies should also include a more fine-grained cognitive–linguistic assessment in order to understand the underlying mechanisms that lead to linguistic errors.

The Contribution of Breath Pauses, Disfluencies, and Mazes to Ratings of Speech Impairment

Another purpose of this study was to examine the extent to which breath pauses, disfluencies, and mazes contribute to ratings of speech impairment. Ratings of speech impairment were significantly related to several factors, including percentage of breath pauses at major syntactic boundaries, percentage of breath pauses at locations unrelated to a syntactic boundary, percentage of breaths at periods, percentage of breaths at locations with no punctuations, number of mazes, and number of disfluencies. However, only two of these variables explained some of the variance in ratings of speech impairment. Together, the number of mazes and the percentage of breath pauses at locations with no punctuation explained 50% of the variance associated with ratings of speech impairment. Speech impairments were rated as more severe in participants who produced more mazes and/or produced more breaths at locations with no punctuation. The fact that speech impairment was rated during a spontaneous speech sample and breath pauses were analyzed during a reading passage could be viewed as a limitation of this study. However, the concordance of the dependent variables from the reading passage with ratings of speech impairment from spontaneous speech strongly suggests that breath pausing patterns and disfluencies impact ratings of speech impairment across speech tasks with different cognitive–linguistic formulation demands.

The link between breath pauses at locations with no punctuation and ratings of speech impairment supports earlier work which demonstrated that listeners use pauses to parse running speech into meaningful units (Price et al., 1991; Shah et al., 2006). When the relationship between breath pauses, syntax, and punctuation is weak, listeners may have difficulty understanding the speaker's message due to errors in how the utterances are parsed. This reduction in understandability is likely reflected in ratings of more severe speech impairment.

On the other hand, the link between ratings of speech impairment and mazes is less established in the dysarthria literature. Hypotheses regarding the variables that contribute to speech impairments have traditionally focused exclusively on speech production variables such as breath pauses. Although overlooked, it makes sense that the ability to formulate complete thoughts and put those thoughts in a logical and concise message would have an impact on the overall impression of speech impairments. Given that individuals, regardless of their level of training, attend to different aspects of speech and voice during perceptual rating tasks (Kreiman et al., 1990), future work should include both speech production and cognitive–linguistic variables when examining the variance in constructs such as speech impairment in individuals with PD.

Clinical Implications

Clinically, these data provide support for the implementation of treatment plans that address both respiratory physiological and cognitive–linguistic systems to drive improved speech outcomes in individuals with PD. Thus, SLPs should conduct comprehensive cognitive–linguistic assessments in addition to traditional motor speech assessments when working with individuals with PD. Examination of breath pause patterns and linguistic errors may be an excellent starting point when forming a treatment plan to address speech impairment. This recommendation is supported by our findings that the production of mazes and the percentage of breath pauses at locations with no punctuation explained half of the variance in ratings of speech impairment over a period of several years.

These data also highlight the benefit of examining breath pausing patterns within the context of a reading passage as opposed to spontaneous speech. Previous work hypothesized that analyses related to syntax and analyses related to punctuation were functionally equivalent (Huber et al., 2012). This study provides support for this hypothesis and further suggests that analyses related to punctuation are more sensitive to measuring change over time and more closely related to ratings of speech impairment. The results for breath pauses at major boundaries were the same as those for breath pauses at periods, and the results for breath pauses at minor boundaries were consistent with those for breath pauses at commas. Results for breath pauses at boundaries unrelated to syntax and for breath pauses at locations with no punctuation were similar at Time 1, but only the measurement of breath pauses at locations with no punctuation resulted in significant differences between the groups at Time 2. Additionally, breath pauses at locations with no punctuation explained a significant portion of variance in ratings of speech impairment. During a motor speech assessment, SLPs can mark their client's breath pauses on a copy of a reading passage and then calculate the percentage of breath pauses taken at periods, commas, and locations with no punctuation. This is a quick and easy objective measure that requires no background knowledge of syntax or instrumental equipment, is likely sensitive to change over time, and is a valid indicator of changes in spontaneous speech following intervention.

Limitations

The primary limitation to this work is the sample size. Longitudinal work is plagued by several challenges including participant attrition and the lack of control of the variables related to disease management, not to mention the significant time and investment needed to conduct the work properly. Small sample sizes in longitudinal work examining a medically complex population are expected. However, future longitudinal research should strive to include more participants. One potential way to handle this difficulty is to establish a nationwide research network dedicated to the longitudinal study of speech and language deficits in individuals with PD. Teams of researchers collecting the same types of data could band together with the intention to develop evidence-based interventions for each stage of the disease.

Another limitation to this work is the subjective nature of auditory–perceptual rating scales, such as the visual analog scale used in this study. Auditory–perceptual ratings can be unreliable, in terms of both intra- and interrater reliability (for a review, see Kreiman et al., 1993). We attempted to correct for interrater reliability by requiring that the difference between the SLP ratings be no greater than 20%. Future work might consider using more listeners or adopting additional criteria for agreement in processing the data.

Conclusions

The current longitudinal study is the first to elucidate the role of disease progression on the coordination of breath pauses with syntax and punctuation as well as the production of speech and linguistic errors in individuals with PD. As the disease progresses, individuals with PD produce fewer pauses at major syntactic boundaries and periods and more pauses at locations with no punctuation. Individuals with PD also produce more disfluencies than control participants. This is also the first study to directly assess and formally establish a relationship between breath pauses, linguistic errors, and ratings of speech impairment. Ratings of the severity of speech impairment were correlated with anumber of breath pause measures and the frequency of disfluencies and mazes. In addition, 50% of the variance in ratings of speech impairment was accounted for by the percentage of breath pauses at locations unrelated to a syntactic boundary and the number of mazes. These results highlight the importance of targeting both respiratory physiological and cognitive–linguistic systems in order to improve speech impairments in individuals with PD. Furthermore, breath pause and linguistic error patterns may be excellent starting points in the treatment of speech impairment due to PD.

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