Research Report

Narrative Discourse Cohesion in Early Stage Parkinson's Disease

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Abstract.

Background: Models of basal ganglia (BG) function suggest that expressive language deficits will likely and consistently present in BG disease. Disparities currently exist between the predictions of models of BG function in expressive language and data from studies of BG disease. Traditional expressive language assessment methodologies that emphasize measures of language form (word and sentence productivity) while not carefully considering how language is used, may only partially account for these disparities.

Objective: To use measures of cohesion to examine the use of cohesive markers in narrative discourse.

Methods: Twelve individuals with idiopathic Parkinson's disease (PD) were compared to 12 matched neurologically intact controls on measures of discourse performance. Three discourse samples (typical day, memorable vacation and family) were analyzed for measures of narrative productivity, number of cohesive ties and cohesive adequacy. Mixed model analyses were completed for group comparisons.

Results: Group differences were not observed on measures of language form as measured by narrative productivity, communication units, and number of cohesive ties produced. In contrast, group differences were observed in cohesive adequacy as individuals with PD produced a higher percentage of incomplete and erroneous cohesive ties relative the control subjects across narratives.

Conclusions: These results support the conclusion that the BG in PD may have an executive role in expressive language use that can be disrupted without impacting language form.

Keywords: Basal ganglia, Parkinson's disease, language, discourse, cohesion

INTRODUCTION

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Parkinson's disease (PD) is a neurodegenerative syndrome that is estimated to affect four to six million individuals worldwide in the world's most populous countries, with 50,000-60,000 new cases being

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diagnosed in the United States annually [1]. While PD affects neuroanatomical regions centering on the basal ganglia (BG), studies by Braak and colleagues have demonstrated that PD related pathology affects multiple neuronal systems as it courses from the brain stem thru the BG and eventually affecting the cerebral cortex [2]. According to Braak et al. [2], the neurodegenerative disease process begins prior to the onset of overt somato-motor dysfunctions. In the earliest stages the disease process begins in the dorsal IX/X motor nucleus and may also extend into the adjoining intermediate reticular zone. The disease then progresses to lower and upper brain stem nuclei followed by an upward course culminating in the cerebral cortex [2]. Because of the extensive disease progression, PD is characterized by progressive reductions in motor as well as cognitive performance.

Models of BG function suggest a critical relationship between the BG and cerebral cortex that should result in predictable expressive language deficits in BG diseases. The BG are connected to the cerebral cortex via a family of cortical-BG-thalamic-cortical circuits that are distinguished by their unique function [3]. Extensive connections exist between most areas of the cerebral cortex, particularly the frontal lobes that are critical to expressive language [4]. Cortical-BG connections provide the anatomical base for the disruption of expressive language which is traditionally linked to the cerebral cortex [5]. Some authors have concluded that the BG and supporting connections contribute to the development of expressive language via word search and generation strategies for verbal expression and therefore expressive language deficits should consistently occur in BG disease [6, 7].

Two recent reviews of language production in PD offer compelling evidence that language disruptions do exist in individuals with PD. Murray's review [8] designed to examine advances in the literature related to changes in language associated with PD offered evidence that language changes in PD existed even in the earliest disease stages. Deficits included morphosyntactic, lexical semantic and language production breakdowns as linguistic complexity increased. A review by Altmann and Troche [9] emphasized the effects of PD on the brain and cognition, and on language production and the stages of language production impairment found as PD progresses. Importantly they described evidence of language production issues following PD that cannot be solely explained by traditional hypotheses of language impairment due primarily to reductions in working memory and/or executive function.

Studies of expressive language associated with BG disease have traditionally emphasized the examination of "language form" (i.e., word and sentence productivity, syntax, grammaticality, etc.) [10–13]. Previous studies have primarily emphasized production at the word and sentence level. Less emphasis has been placed on how language is being used (language use), particularly at the discourse or conversational level. These issues are important because successful generation of expressive language involves the development of language form and the correct use of that form. As a result, studies designed to examine language use in those with BG disease are needed to establish the hypothesized vulnerability of expressive language use in BG disease. However, approaches designed to study language use and language form have yet to emerge. One method for making this comparison may be to analyze discourse for attributes of language form and use during discourse production. The assessment of discourse can be used to evaluate the complex integration of sentences into coherent communication that represents the speaker's skills in language use and relating language use to the grammatical form [14]. In summary, discourse contains attributes of language form and language use that can be measured in one sample.

Narrative discourse is used to describe an experience as a sequence of events or episodes [15]. Successful narrative discourse contains the linking of meaning across sentences or cohesion. Cohesion occurs via the use of cohesive markers that direct the listener to information found outside individual sentences [16]. A cohesive marker creates a tie with information found outside individual sentences, thus establishing a meaningful relationship across sentences [17]. Cohesion has been measured in disorders that are known for language use deficits; Alzheimer's disease [18], traumatic brain injury [19] and stroke [20]. Because sentence production involves a complex sequence of choosing lexical items and appropriate sentence structures, disease of the BG may disrupt the language planning process required to produce fluent and cohesive output [10].

Therefore, the purpose of this study is to use measures of cohesion in narrative discourse to examine language use in individuals with BG disease. We selected individuals with idiopathic PD because they have a disease that begins in the BG and disrupts the cortical-BG circuits that govern various aspects of higher cortical functioning [21]. We hypothesize that individuals with PD would exhibit differences in the cohesiveness of their narratives relative to neurologically intact controls as a result of BG disease and disruptions of cortical-BG functions. We elected to test this hypothesis in individuals with early stage PD because disease related pathology exists even during asymptomatic preclinical phases before the overt signs of the disease emerge [22].

MATERIALS AND METHODS

Description of the subjects

Participants consisted of 12 individuals diagnosed with idiopathic PD (hereafter referred to as experimental subjects) by a movement disorders neurologist using the strict criteria of the UK Brain Bank [23] and 12 individuals who were age, education, ethnicity and gender matched and neurologically intact (hereafter referred to as control subjects). The study was IRB approved and all participants gave written informed consent. All participants were male, right handed, and had no history of prior stroke, dementia, brain tumor, or head trauma. All had at least a seventh grade education, functional hearing for normal conversation, functional vision for reading tasks, spoke English as their primary language, and demonstrated expressive language skills within intact range for normal conversation. All subjects (experimental and control) exhibited scores of 26 or better on the Mini Mental Status Exam (MMSE) [24].

Each experimental subject presented with a minimum of 3 of 4 cardinal features of PD (resting tremor, rigidity, bradykinesea, postural instability) and had no history of deep brain stimulation or brain lesion therapy. The parkinsonism of each experimental subject was rated with the Hoehn & Yahr Staging Scale for PD and classified by predominate feature (tremor vs. rigidity).

Procedure

Standardized assessments

The Boston Naming Test (BNT) [25] and Wechsler Memory Scale – Logical Memory I (WMS-LMI) [26] were administered to examine potential group differences relative to language form (BNT) and the influence of memory on language form and use (WMS-LMI).

Narrative discourse data collection

Narratives were collected from experimental subjects in the "off" state of their parkinsonian medications. Experimental subjects were seen in their homes prior to their first daily dose of anti-parkinsonian medication and at least 12 hours since their last dosage to maximize dopamine depletion. Five of the 12 experimental subjects were newly diagnosed with PD and had no history of parkinsonian medication use at the time of the study. Control subjects were seen primarily in their homes.

Each participant was instructed to discuss 3 topics, which included the following: (1) a typical day, (2) a memorable vacation, and (3) his family for a minimum of 3-minutes. In the event that any subject stopped before 3-minutes, a standardized verbal cue was provided to continue the narrative until the 3-minute minimum was achieved. The order of presentation of the 3 topics was counterbalanced across subjects. All responses were audio recorded for off line transcription and scoring.

Analysis of narrative data

Transcription and segmentation The first 3 minutes of all language samples were transcribed verbatim. Each sample was divided into communication units (CU); defined as the shortest allowable independent clause and related dependent clauses. Individual CU's were defined primarily by syntax, however prosodic and semantic features were used at times when the unit could not be determined entirely by syntax. All words unintelligible to the investigator were excluded from the analysis. In instances where the location of coordinating conjunctions such as "and", "but" and "or" was unclear, their prosodic feature determined their final location at the beginning or ending of the communication unit. One-word responses were not considered in the communication unit calculation. Scoring guidelines for communication units were based on Hunt's procedure [27].

Calculation of communication units, words, and cohesive ties The total number of CU's and words in each transcribed narrative were calculated for comparisons. Following segmentation of CU's, the total number of communication units and words for the 3-minute sample was calculated for each subject. Each communication unit was then evaluated for use of cohesive ties. Cohesive ties within three categories (Reference, Conjunction, and Lexical) were identified. Although five types of cohesive ties exist, studies of discourse cohesion report that typically only three ties are used frequently enough for statistical analysis [28] (see supplemental material for definitions and discourse examples). *Calculation of cohesive adequacy* Each cohesive tie was judged for the adequacy of its tie (complete, incomplete or erroneous). Cohesive ties were judged "complete" when the referent could be easily found in the preceding discourse. Ties were defined "incomplete" cohesive markers when the referent could not be identified in the preceding discourse or was not evident. Ties were defined as "erroneous" when multiple referents could be identified in the discourse therefore making the marker ambiguous.

The percentage of incomplete and erroneous ties were calculated as defined below.

% incomplete/erroneous ties

 $=\frac{total \ incomplete \ and \ erroneous}{ties \ total \ cohesive \ ties}$

Scoring reliability Three trained raters participated in the project to establish reliability for identification of CU's, words, number of cohesive ties and percent incorrect and erroneous use of cohesive ties. Raters were blinded to the neurological status subjects that generated the samples used for the analyses. One trained rater analyzed 100% of the samples that were used for the analysis. Two additional trained raters independently analyzed 15% of the total sample. Intra-class correlation coefficients were completed to analyze agreement among the 3 raters.

Analysis of motor speech performance

An independent judge blinded to the neurological status of all subjects rated motor speech performance of all audio recorded samples. Each sample was rated on a 5-point scale of speech intelligibility. Ratings ranged from 1 (No detectable disorder) to 5 (No functional speech). Speech ratings were correlated with all language variables.

RESULTS

Scoring reliability

Intra-class correlation coefficients (ICC) were calculated by using a two-way mixed model with repeated measures to evaluate scoring agreement among the raters for CU's, words, cohesive ties and cohesive adequacy. ICC scores of (0.99) were achieved for CUs and words, (0.91) for cohesive ties and (0.83) for percent correct use of ties.

Statistical analyses

Mixed model analyses (2 group \times 3 narrative topics; (<0.05) were completed for all analyses unless otherwise stated. The examination of CU's, total words, cohesive ties, and cohesive adequacy compared only the first 3-minutes of each narrative sample.

Demographic comparisons

A detailed profile of the PD subjects is provided in Table 1. Two-tailed *t*-test, alpha level = 0.05, revealed no significant group differences between the experimental and control subjects on age, education, and standardized cognitive and language comparisons (see Table 2).

	Table 1												
Demographic, cognitive and language characteristics of PD subjects													
ID	Age	Education	Parkinson	H&Y	BNT	MMSE	WMS-						
	(Years)	(Years)	Years				LMI						
P1	85	12	0	2	53	26	12						
P2	74	12	0	3	52	30	43						
P3	84	11	4	3	53	29	9						
P4	78	12	7	3	51	29	37						
P5	64	14	0.5	2	60	27	36						
P6	84	12	0	2	33	26	13						
P7	69	14	0	2	56	30	25						
P8	61	12	9	3	55	29	28						
P9	76	12	14	3	56	29	38						
P10	83	9	0	2	50	30	34						
P11	64	12	7	2	58	29	20						
P12	40	12	2	2	56	29	35						

Parkinson Years = the number of years since PD subjects were initially diagnosed with PD. H & Y=Hoehn and Yahr. BNT = Boston Naming Test, all items administered. MMSE = Mini Mental Status Exam. WMS-LMI = Wechsler Memory Scale – Logical Memory I subtest.

 Table 2

 Demographic, cognitive, and language comparisons

Variable	PD subjects		Controls		Т	р
	М	SD	М	SD		
Age (years)	71.8	13.2	72.6	13.5	-0.14	>0.05
Education (years)	12.0	1.3	12.8	2.8	-0.94	>0.05
Parkinson Years	3.6	4.6				
H & Y stage	2.4	0.5				
BNT	52.8	6.7	51.8	8.4	0.31	>0.05
MMSE	28.6	1.4	28.8	1.7	-0.26	>0.05
WMS-LMI	27.5	11.5	30.6	144	-0.58	>0.05

p values are derived from comparisons of PD subjects to normal controls. Parkinson Years = the number of years since PD subjects were initially diagnosed with PD. H & Y = Hoehn and Yahr. BNT = Boston Naming Test, all items administered. MMSE = Mini Mental Status Exam. WMS-LMI = Wechsler Memory Scale – Logical Memory I subtest.

406

70

68

66

64

62

Language form

Communication units and words

Results of a 2 (group) \times 3 (narrative) comparison indicated non-significant effect for group on total number of CU's [F(1, 23) = 0.99, p = 0.33] and total number of words [F(1, 23) = 0.00, p = 0.98). The experimental subjects produced similar numbers of CU's and words relative to the controls across the 3 narratives (typical day, memorable vacation, and family). A group \times narrative interaction was significant for CU's [F(2, 192) = 3.90, p = 0.022]. Pairwise comparisons using independent sample *t*-tests indicated that controls subjects exhibited a significant reduction in CU's during their discussion of family relative to vacation (t = 2.865; p = 0.015).

Language use

Cohesive ties

Results of a 2 (group) \times 3 (narrative) comparison indicated a non-significant effect for group [F(1,24) = 0.010, p = 0.919]. The experimental subjects produced similar numbers of total cohesive ties relative to the controls across the 3 narratives (typical day, memorable vacation, and family). A group \times narrative interaction was significant [F(2, 192) = 10.48,p = 0.000]. Pairwise comparisons indicated that control subjects produced significantly fewer cohesive ties during their discussion of typical day relative to memorable vacation (t = -2.314; p = 0.04). The mean number of cohesive ties produced by the 2 groups is reported in Fig. 1. A percentage distribution of each cohesive tie type (Reference, Lexical, and Conjunction) was calculated and is displayed in Fig. 2.

Cohesive adequacy

Results of a 2 (group) \times 3 (narrative) comparison indicated a significant main effect for group [F(1, 81) = 5.891, p = 0.017]. The experimental subjects produced a higher percentage of incomplete and erroneous cohesive ties relative to the control subjects across narratives. There were no significant group \times narrative interactions. The mean percentage use of incorrect and erroneous of cohesive ties produced by the 2 groups is displayed in Fig. 3.

Further inspection of the cohesive ties judged as incomplete or erroneous indicated that the experimental subjects produced 136 ties identified as incomplete or in error compared to 63 by the controls subjects. Sixty-nine percent of all ties were judged incomplete or



Cohesive Ties

*

Fig. 1. Mean number of cohesive ties for each narrative. *Controls produced fewer cohesive ties during typical day compared to vacation



Fig. 2. Percentage distribution of each cohesive tie type.



Incomplete & Erroneous Cohesive Ties

Fig. 3. Percent Incomplete and Erroneous Cohesive Ties. *PD subjects produced higher percentage of incomplete and erroneous ties.

erroneous were of reference type for both groups. The total number of ties judged incomplete or erroneous is depicted in Fig. 4.



Incomplete and Erroneous Ties

Fig. 4. Number of incomplete or erroneous ties for each narrative.

Motor speech performance

A two-tailed *t*-test (p < 0.05) revealed a significant group difference between the experimental subjects (M=2.2, SD 0.72) and control subjects (M=1.3, SD 0.62) on intelligibility ratings, t (22), p=0.003. Correlations with language variables indicated nonsignificant correlations between ratings of motor speech performance and CUs (r=0.19, p=0.38), words (r=-0.07, p=0.76), total ties (r=0.05, p=0.81), and percent incomplete and erroneous cohesive ties (r=-0.21, p=0.33).

DISCUSSION

The results of this study confirmed a disorder of language use (cohesive adequacy) in those with early PD even though significant differences were not observed on measures of language "form" or by motor speech issues. Relative to the controls, the experimental subjects produced similar language form as measured by number of: CU's, words and cohesive ties. In contrast, individuals with PD produced a greater percentage of incomplete and erroneous ties in their narratives. Thus, while aspects of overall "form" of narrative discourse output (CU's, total words, total cohesive ties) may be relatively preserved in the earliest stage of PD, the cohesiveness of narrative discourse may be impaired, heralding the emergence of language use disruption.

These findings also support the notion that traditional measures of language "form" and measures designed to capture language production issues at the sentence level may be insensitive to language disruption in the earliest stages of PD. It is presumed that to adequately understand language production among individuals with diseases of the BG such as PD, studies should be designed to emphasize "language use" at a more complex level than word production tasks, i.e., discourse. This presumption is supported by studies which have shown impairments in language pragmatics or the use of verbal and non-verbal social communication in individuals with PD [29].

In this study, a review of all incomplete or erroneous ties revealed that experimental subjects produced double the number of incomplete or erroneous ties compared to the control subjects. Sixty nine-percent of all ties judged as incomplete or in error were of the reference type for both groups. Reference is reportedly susceptible to disruption in neurological disease due to the complexity of the reference system. The use of reference is important to narrative discourse as it directs the listener to the identity of the thing to which the references refers [16].

Our observed greater number of incomplete and erroneous cohesive ties among individuals with PD compared to controls agree with previous studies of cohesion in neurological disease [18, 30]. Ripich and Terrell [31] observed significantly greater use of incomplete and erroneous cohesive ties among individuals with dementia compared to elderly controls. They hypothesized that reference ties serve to structure or organize narrative discourse production and may be more sensitive to neurological disease. The lack of equivalent reductions in cohesive adequacy among lexical and conjunction ties suggests that general nouns and conjunctions create less opportunity for ambiguity relative to pronouns. Consequently, use of reference ties may be more subject to increased inaccuracy of use relative to lexical and conjunction ties in PD. It is important to note that we only saw the participants in the "off" medication state. Completing the same measures during their "on" state may have offered additional information regarding the impact of PD on cohesiveness during discourse production. Similarly, a more comprehensive measure of cognitive functioning with a particular emphasis on executive function would ensure that the observed language errors were independent of other cognitive issues.

Cohesion is a microlinguistic feature of narrative discourse that has the potential to be masked in PD by more commonly observed and reported motor speech deficits. Van Leer & Turskstra [28] noted that ambiguous intersentential meaning can occur when the cohesive tie is not readily apparent. Reductions in cohesion are believed to result in increased vagueness thereby confusing the intended listener [18]. Glosser and Deser [31] also reported that disrupted cohesion can reflect impaired lexical retrieval during discourse construction. Since cohesion serves to facilitate the continuity of meaning in narrative discourse, it is believed to provide an indirect index of the ability to maintain a topic during discourse production. Therefore, disruptions in cohesive adequacy can indicate a more global disruption of language use in PD.

There is current evidence related to the neurobiological underpinnings of a potential increase in incomplete/erroneous cohesive ties in PD. For example Ford et al. [32] demonstrated that the machinery for basal ganglia participation in language function exists by demonstrating connections between the basal ganglia and Broca's area and as previously hypothesized by Ullman [33]. However, simple connectivity does not explain the role of the basal ganglia in discourse processing. But, more comprehensive neurocognitive models designed to address linguistic functions of the basal ganglia for word or sentence processing have application for the interpretation of the current discourse data. Evidence is offered from three sources. First, Crosson et al. [34] addressed the role of a left pre-SMA-basal ganglia loop in word production. The authors hypothesized that the basal ganglia increases the signal-to-noise ratio during word selection by enhancing activation of the best lexical candidate while suppressing other competing candidates. More specifically, the best candidate selection in discourse is determined by the surrounding context as well as by semantic considerations. According to this theory, incomplete or erroneous cohesive markers would be selected due a failure to enhance an appropriate choice over less appropriate ones. For example, a pronoun (reference tie) with multiple equally possible prior referents could be selected instead of a word making the reference to the prior concept more specific because of a failure to enhance activation of the specific word more than that of the non-specific pronoun (which was common in our PD discourse samples). The biggest weakness in this explanation is that one would anticipate other word selection difficulties, which typically are not considered to be prominent in PD.

A second potential explanation for more incomplete/erroneous cohesion ties involves automatic processes, in particular procedural memory. Studies by Copland et al. have suggested that lexical-semantic processing difficulties in PD occur in automatic as opposed top-down processing [35, 36]. Support for that explanation emerged from Ullman who suggested the basal ganglia plays a prominent role in the procedural memory system underpinning grammatical functions [33]. In keeping with a vast literature on procedural memory, Ullman defines procedural memory as subserving "the learning of new, and control of established sensory-motor and cognitive 'habits', 'skills', and other procedures" [33, p. 237]. Ullman's concept is consistent with the observations of Copland in that procedural memory is capable of operating outside of top-down control mechanisms. This explanation assumes that discourse cohesion is controlled, at least in part, by procedural memory systems affected by PD. However, our data indicate that the overall "form" of narrative discourse (CU's, total words, total cohesive ties), the mostly likely aspect of discourse to be influenced by procedural memory, was relatively intact.

The third explanation for greater use of incomplete/erroneous cohesion ties during discourse production in PD involves the role or working memory in cohesion for discourse. Working memory is the ability to hold information in one's short-term memory for use or manipulation during ongoing cognitive operations. In that regard, Grossman et al. concluded that working memory limitations played a role in the difficulties that PD patients have with complex syntax and that these working memory limitations were due to frontal-basal ganglia dysfunction in PD [37, 38]. In this explanation, incomplete or erroneous cohesive ties would be related to PD patients losing track of all of the prior information in their narrative to which cohesive ties might be made. Consequently, a PD

patient might select a pronoun with multiple potential referents because s/he has lost the ability to track all of the potential referents. Future studies must be designed to test the working memory scenario against the aforementioned scenarios by including more detailed measures of working memory, procedural memory, and word retrieval in a study of discourse beyond the measures used in this study to determine which function(s) are most strongly associated with increases in incomplete/erroneous cohesive ties.

Though many questions persist regarding the influences of BG disease on expressive language performance, our findings indicate that language use deficits do in fact occur. While individuals with BG disease may exhibit fluent verbal output, the cohesiveness of their expressive language may be deceased. It is important that we acknowledge the potential contributions to discourse production from the diversity of cortical and subcortical structures identified by Braak and colleagues that are involved in the PD disease progression [2]. Novel methodologies will be required to test the sensitivity of BG disease to expressive language performance that address potential language use deficits such as correct pragmatic language use and the relative contributions from extra-BG anatomical structures to language use issues. Cohesion analyses with special emphasis on cohesive adequacy can provide a useful means to identify sensitive expressive language impairments that will not be otherwise identified. Similarly, clearly understanding the exact nature of cohesion issues may lead to interventions that parallel motor speech treatments that are designed to reduce the impact on communication and minimize the progressive impact of the disease. Finally, the observation of group by narrative interactions on measures of CU's and total cohesive ties suggests that multiple samples are required to adequately consider the variability of performance during discourse production when examining language form and language use variables.

CONCLUSIONS

Studies of individuals with disease of the BG have generally emphasized motor speech and language form changes while omitting expressive language use issues. Models of BG functioning suggesting that BG diseases should have a greater influence on expressive language use were supported by the findings of this study.

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SUPPLEMENTARY MATERIAL

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