Research Article

Internally Versus Externally Cued Speech in Parkinson's Disease and Cerebellar Disease

Phil Weir-Mayta,^a Kristie A. Spencer,^b Tanya L. Eadie,^b Kathryn Yorkston,^c
Sara Savaglio,^b and Chris Woollcott^b

Purpose: The purpose of this study was to examine the effects of an internally versus externally cued speech task on perceived understandability and naturalness in speakers with Parkinson's disease (PD) and cerebellar disease (CD).

Method: Sentences extracted from a covertly recorded conversation (internally cued) were compared to the same sentences read aloud (externally cued) by speakers with PD and a clinical comparison group of speakers with CD. Experienced listeners rated the speech samples using a visual analog scale for the perceptual dimensions of understandability and naturalness.

Results: Results suggest that experienced listeners rated the speech of participants with PD as significantly more

natural and more understandable during the reading condition. Participants with CD were also rated as significantly more understandable during the reading condition, but ratings of naturalness did not differ between conversation and reading.

Conclusions: Speech tasks can have a pronounced impact on perceived speech patterns. For individuals with PD, both understandability and naturalness can improve during reading tasks versus conversational tasks. The speech benefits from reading may be attributed to several mechanisms, including possible improvement from an externally cued speech task. These findings have implications for speech task selection in evaluating individuals with dysarthria.

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It is well established that individuals with Parkinson's disease (PD) have difficulty performing skilled movement when forced to rely on internal cues versus responses guided by external cues or stimuli, such as a line on the floor to guide gait patterns (Gowen & Miall, 2007; Ketcham, Hodgson, Kennard, & Stelmach, 2003; Majsak, Kaminski, Gentile, & Flanagan, 1998; Morris, 2000; Sawamoto, Honda, Hanakawa, Fukuyama, & Shibasaki, 2002; Van Donkelaar et al., 2000). Most attribute this difficulty to dopaminergic deficits affecting the basal ganglia and fronto-striatal networks, which are involved in the preparation of internally generated movements (Cunnington, Iansek, Bradshaw, &

Phillips, 1995; Lewis, Byblow, & Walt, 2000; Morris, 2000; Schenk, Baur, Steude, & Bötzel, 2003). As a result, individuals with PD become overly reliant on visual/auditory external stimuli to guide skilled movement (Romero, Van Gemmert, Adler, Bekkering, & Stelmach, 2003). Converging evidence suggests marked improvement in performance with external cueing for behaviors such as walking (Luessi, Mueller, Breimhorst, & Vogt, 2012; Mak, Yu, & Hui-Chan, 2013), writing (Oliveira, Gurd, Nixon, Marshall, & Passingham, 1997; Teulings, Contreras-Vidal, Stelmach, & Adler, 1997), and driving (Scally et al., 2011; Stolwyk, Triggs, Charlton, Lansek, & Bradshaw, 2005).

Internal Versus External Cueing and Speech

Evidence is also emerging regarding the influence of internal versus external cueing on speech production. The benefits of external cues have been demonstrated in broader clinical contexts and, most germane to this study, during specific speech tasks. To highlight, in a clinically oriented study of pacing and speech rate, speakers with PD and severe hypokinetic dysarthria significantly improved overall mean intelligibility in the presence of rhythmic auditory cues (Thaut et al., 2002). The authors speculated that the rhythmic auditory cues acted as pace keepers for the impaired

^aDepartment of Communication Sciences and Disorders, California State University, Fullerton

^bDepartment of Speech and Hearing Sciences, University of Washington, Seattle

^cDepartment of Rehabilitation Medicine, University of Washington, Seattle

Correspondence to Phil Weir-Mayta: pweir-mayta@fullerton.edu

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internal model. This finding parallels the use of rhythmic auditory cues to improve festinating gait in individuals with PD (Lim et al., 2005).

In addition to rhythmic auditory cues, benefits of external cueing in speakers with PD is also evident in the speech volume regulation literature. Ho, Bradshaw, Iansek, and Alfredson (1999) found that individuals with PD were able to successfully adjust their speech volume when provided with verbal (instructional) cues to focus on their loudness, but not with implicit cues integral to volumetric scaling. The ability of speakers with PD to regulate their volume through the use of external cues has also been robustly demonstrated by research on the Lee Silverman Voice Treatment (Ramig, Pawlas, & Countryman, 1995), which teaches hypophonic speakers to disregard their defective internal cues and instead “recalibrate” their volume using external cues to “think loud” (Fox et al., 2006).

Studies specifically examining speech tasks have often designated reading as an externally cued condition. Although printed text is not considered a cue in the traditional sense, it may similarly serve as an external trigger/stimulus, and lead speakers with PD to favorably alter the understandability and/or prosodic aspects of speech. For example, seeing the spacing between the printed words might trigger a speaker to provide more distinct word boundaries, or might lead the speaker to say the sentence more fluently or more loudly. Further, syntactic markers such as commas and periods are known to influence aspects of prosody, such as breath patterning, intonational contours, and rate in both healthy adult speakers and speakers with PD (Huber, Darling, Francis, & Zhang, 2012; MacPherson, Huber, & Snow, 2011; Steinhauer, 2003). Thus, written text may serve as an external cue to enhance performance similar to findings from the limb motor control literature.

There have been several comparisons of internally versus externally cued speaking conditions in individuals with PD. Kempler and Van Lancker (2002) conducted a single-case study evaluating intelligibility across five speech tasks, including monologue and reading. Results showed that the speaker had significantly better intelligibility (78%) for the reading task versus the monologue task (29%). The authors attributed the low intelligibility of the monologue to the inability of the basal ganglia to provide an accurate internally generated model. The reading and repeating tasks putatively provided enough external support to reduce the demands on the basal ganglia for planning, initiating, and/or monitoring speech gestures. In a similar single-case study of a man with severely dysfluent speech associated with parkinsonian syndrome, Van Lancker Sidtis, Cameron, and Sidtis (2012) reported that spontaneous speech was more impaired on fluency outcome measures than the externally cued tasks of repetition, reading, sentence repetition, and singing.

In a similar manner, Sidtis, Cameron, Bonura, and Sidtis (2012) asked six individuals with PD and five individuals with PD plus deep brain stimulation (DBS), all with mild hypokinetic dysarthria, to provide speech samples in two tasks—monologue and repetition of the monologue utterances. Listeners rated speech intelligibility and difficulty

in listening to the speech using a 5-point rating scale. There were significant effects of task and DBS on intelligibility measures of speech, with similar findings for the difficulty ratings. Repetition was more intelligible than conversation, leading the authors to suggest a facilitative role of external models provided by repetition in motor speech performance.

Other researchers have found questionable benefit to speech from the provision of external cues. For instance, Bunton and Keintz (2008) calculated speech intelligibility across a number of different speech tasks in single- and dual-task conditions for four speakers with hypokinetic dysarthria from PD and four age-matched controls. Participants provided three types of speech samples: (a) reading monosyllabic words (external cue), (b) reading low-predictability sentences (external cue), and (c) producing a monologue about a recent vacation (internal cue), in both single- and dual-task conditions. In addition to the speech tasks above, researchers covertly recorded a 2–3-min spontaneous speech sample, obtained by asking the participant open-ended questions to encourage conversation. Listeners provided intelligibility measures across the tasks.

In the single-task condition (speaking only), intelligibility for both control participants and participants with PD was similar for the three overt speech tasks, with no significant differences noted between externally cued tasks (reading single words, reading sentences) and the internally cued task (monologue). For controls, the difference between scores from the overt monologue and the *conversational* spontaneous speech sample was not statistically significant. However, for participants with PD, the difference in intelligibility between the overt monologue and the conversational spontaneous speech sample (both internally cued) was statistically significant. The authors hypothesized that intelligibility was better during monologue production versus spontaneous speech due to the participant’s awareness of the recording, resulting in additional attentional focus on the task. This finding is consistent with previous reports of performance effects in speakers with PD (Keintz, Bunton, & Hoit, 2007; Sarno, 1968; Weismer, 1984). Taken together, these findings implicate attention as a possible causal factor in increased performance on speaking tasks, and suggest negligible benefit of external cues for speech production in individuals with PD.

A study by Tjaden and Wilding (2011) also indicated questionable benefit from provision of external cues in speech tasks on the basis of 12 participants with PD (10 with hypokinetic dysarthria, one with hyperkinetic dysarthria, and one with hypo-hyperkinetic dysarthria). Intelligibility estimates were obtained for a reading passage and an extemporaneous 2-min monologue. Sixty untrained listeners judged intelligibility for the reading passage using orthographic transcription and modulus-free direct magnitude estimation (DME). For DME rating, intelligibility was operationally defined as “the ease with which speech is understood.” Analysis of scaled scores generated by DME indicated no significant difference in scaled intelligibility for the paragraph reading and monologue tasks. The authors subsequently cautioned against the assumption that findings from the general motor

control literature might extend to speech in PD. However, the authors also cited anecdotal evidence from a previous study (Tjaden & Wilding, 2004) conveying that listeners find the DME task of numerically scaling intelligibility, in the absence of an anchor or modulus, to be unusual (Tjaden & Wilding, 2011). This may explain why measures of listener reliability for DME were not overwhelmingly strong in the 2011 study (Reading Passage: $M = 0.33$ and Monologue: $M = 0.42$).

Together, findings from Tjaden and Wilding (2011), as well as Bunton and Keintz (2008), demonstrated comparable intelligibility for internally and externally cued speech tasks. These outcomes differ from reports indicating poorer intelligibility for monologue in PD, compared with tasks such as reading words and sentences (Canter & Van Lancker, 1985; Kempler & Van Lancker, 2002; Sidtis et al., 2012). Their findings also differ from limb motor control studies reporting that externally cued movements for individuals with PD are superior to self-initiated movements (Burleigh-Jacobs, Horak, Nutt, & Obeso, 1997; Cunnington, Iansek, & Bradshaw, 1999; Lehman, Toole, Lofald, & Hirsch, 2005; McIntosh, Brown, Rice, & Thaut, 1997; Morris & Iansek, 1996; Thaut et al., 1996). These discrepancies may be due, in part, to differences in methodology (e.g., rating scales vs. transcription of utterances; the experience level of the listeners, etc.) and outcome measures, making it difficult to draw conclusions about the influence of internally versus externally guided speech tasks. It is also possible that reading tasks may not serve as external cues and thus provide no benefit to speech productions, unlike the benefits seen in the limb movement literature.

Naturalness as an Outcome Measure

Although intelligibility is often affected in speakers with PD, it more strongly reflects articulatory function than alterations to prosody (Anand & Stepp, 2015). Moreover, it is quite typical for speakers with PD to have relatively preserved intelligibility, but disrupted prosody (Duffy, 2013). Yet to date, studies of speaking task differences in PD have focused primarily on speech intelligibility, often overlooking important contributing factors such as naturalness (see, however, Anand & Stepp, 2015; Sussman & Tjaden, 2012). Speech naturalness relates to a speaker's *prosody*, which encompasses speech rate, rhythm, intonation, stress patterns, and loudness (Yorkston, Beukelman, Strand, & Bell, 1999)—features that are often dramatically and pervasively affected in speakers with dysarthria from PD. Thus, relying solely on intelligibility may not provide the most sensitive or complete indicator of speech changes across internally versus externally guided cueing conditions.

Naturalness is important to study in speakers with PD because of the pronounced psychosocial ramifications. Studies have shown that dysprosody from PD contributes to difficulty expressing emotions (Pell, Cheang, & Leonard, 2006) and difficulty conveying meaningful distinctions in communication (Pell & Leonard, 2003). Dysprosody can also affect social impressions of individuals with PD (Pitcairn,

Clemie, Gray, & Pentland, 1990). For example, in a study by Jaywant and Pell (2010), speakers with PD were perceived as significantly less interested, less involved, less happy, and less friendly than healthy speakers. Thus, speech naturalness is an important outcome measure to implement and understand in speakers with PD.

Use of a Clinical Control Group

To determine the specificity of an external cueing effect, speakers with hypokinetic dysarthria will be compared to speakers with ataxic dysarthria from cerebellar disease (CD) in the present study. Although individuals with PD have “scaled down” motor output that external cues may help to overcome, this pattern does not hold for individuals with CD. In the case of CD, the motor coordination and regulation impairment from the ataxia should be present regardless of context (Cannito & Marquardt, 1997; Duffy, 2013; Kent, Kent, Weismer, & Duffy, 2000). Thus, external cueing would not be anticipated to influence the speech of participants with ataxic dysarthria. Speech improvement with external cueing for the speakers with hypokinetic, but not ataxic, dysarthria would support the premise that the dysfunctional basal ganglia is lacking an adequate, internally generated model, and speakers with PD become reliant on external cues to guide skilled movement.

In sum, a review of the speech literature on internally versus externally cued tasks in speakers with PD reveals an unclear picture. Although some investigators suggest intelligibility is negligibly changed when an external auditory or visual cue is provided (Bunton & Keintz, 2008; Tjaden & Wilding, 2011), others suggest that individuals with PD speak more intelligibly during speech tasks that provide an external model (Kempler & Van Lancker, 2002; Sidtis et al., 2012). Results are difficult to reconcile, in part due to differences in methodology. Moreover, studies to date have maintained a narrow focus on intelligibility as the primary outcome measure, and have not implemented a clinical control group to determine the specificity of external cueing effects. To address this gap in research, the current study expanded the outcome measures to capture subtle changes in speech that can be masked solely by measuring intelligibility. The expanded measures include understandability and naturalness, with the intent of focusing on articulation and prosody, respectively. To further understand speech task differences, speakers with ataxic dysarthria were included as a clinical comparison group. We hypothesized that understandability and naturalness would be rated more favorably in the externally cued speech task in the participants with PD. This finding would support the hypothesis that the dysfunctional basal ganglia are lacking an adequate, internally generated model that integrates sensory input with motor output to guide movements (including speech). Thus, external cues may improve movement through the use of increased sensory input, in turn helping to guide motor output. As a result, speakers with PD become reliant on external cues to guide skilled movement. It is also possible that differences between reading

and covert speech are related to participants “performing” the task; this would represent a simple change in behavior, as much as a broad neurophysiologic change, and would be expected to manifest in both groups.

Method

Participants

The Institutional Review Board of the University of Washington approved this study. All participants in the study provided written, voluntary, informed consent.

Speakers

A total of 16 adults completed the study; a summary is provided in Table 1. Ten were diagnosed with PD (nine men and one woman) and ranged in age from 59–78 years ($M = 69.7$, $SD = 5.5$). Six were diagnosed with CD (one man and five women) and ranged in age from 41–73 years ($M = 58.5$, $SD = 11.4$). All participants were native speakers of American English and had adequate visual acuity demonstrated by accurately reading line 20/30 at 2.3 ft from the Snellen chart. Adequate hearing thresholds in at least one ear ≤ 50 dB at 500, 1000, and 2000 Hz was also a requirement. Typical speech, language, and cognitive developmental history was also a requirement for inclusion per self-report. All participants reported a neurologist PD or CD diagnosis (per self-report). Participants continued taking their prescribed medications during the experiment. To minimize the impact of medication use, speakers were scheduled during their optimal medication period (1–2 hr after medication, per self-report). Participants

were excluded for prior neurosurgical procedures, including DBS. All participants were at least 5 years post diagnosis ($M = 10.6$ years, $SD = 6.2$) and were required to successfully pass a depression and cognitive screening. To be specific, participants needed to score ≤ 13 on the Beck Depression Inventory II (Beck, Steer, & Brown, 1996) and ≥ 25 on the Mini-Mental Status Exam (Folstein, Folstein, & McHugh, 1975). No participants ultimately were excluded on the basis of these screening measures. However, out of 21 participants originally recruited for the study, five participants with PD were excluded for not having a discernible dysarthria, thus yielding a final count of 16 eligible participants.

Presence and severity of dysarthria was verified by two of the authors (K.A.S. and K.M.Y.) who are both speech-language pathologists and researchers, each with more than 20 years of experience with dysarthria. The expert listeners heard a sample of connected, spontaneous speech from each participant that had been recorded, digitized at a 44000 Hz sampling rate, and presented in a quiet room using Adobe Audition 9.0 (Adobe Systems Inc., San Jose, CA). Judgments of speech understandability and naturalness were made on a 4-point ordinal scale (i.e., *normal*, *mild*, *moderate*, and *severe*), with expert raters reaching 100% agreement on all samples. In cases where mixed dysarthria was possible (i.e., ataxic with spastic), it was agreed that the predominant dysarthria was hypokinetic or ataxic.

Listeners

Ten licensed speech-language pathologists with a minimum of 5 years of clinical experience (Bunton, Kent,

Table 1. Characteristics of participants with Parkinson’s disease (PD) and cerebellar disease (CD).

Participant	Gender	Age	Years of education	Etiology	Dysarthria type	Years since diagnosis	Speech intelligibility test (Yorkston et al., 2007)		
							Percent intelligible	Severity level - U	Severity level - N
PD1	M	74	20	PD	Hypokinetic	18	96	MOD	MOD
PD2	M	65	16	PD	Hypokinetic	8	97	MILD	MILD
PD3	M	72	16	PD	Hypokinetic	8	96	MOD	MILD
PD4	F	59	16	PD	Hypokinetic (+ spastic?)	20	93	MILD	MILD
PD5	M	73	16	PD	Hypokinetic	10	89	MOD	MOD
PD6	M	71	19	PD	Hypokinetic	10	45	SEV	SEV
PD7	M	64	18	PD	Hypokinetic	5	82	SEV	SEV
PD8	M	70	18	PD	Hypokinetic	5	98	MILD	MOD
PD9	M	78	18	PD	Hypokinetic	7	97	MILD	MILD
PD10	M	73	18	PD	Hypokinetic	5	91	MOD	MILD
<i>M</i>	9 M, 1 F	69.7	17.5			9.6	88.4		
<i>SD</i>		5.5	1.4			5.3	16.0		
CD1	F	60	17	CB stroke	Ataxic	6	95	MILD	MILD
CD2	M	73	12	SCA6	Ataxic	20	77	SEV	MOD
CD3	F	41	18	Friedreich’s ataxia	Ataxic	22	94	MILD	MILD
CD4	F	53	14	SCA3	Ataxic	11	99	MILD	MILD
CD5	F	68	18	Progressive CB ataxia	Ataxic	2	97	MILD	MOD
CD6	F	56	14	Progressive CB ataxia	Ataxic (+ spastic?)	12	97	MILD	MOD
<i>M</i>	1 M, 5 F	58.5	15.5			12.2	93.2		
<i>SD</i>		11.4	2.5			7.8	8.1		

Note. U = understandability; N = naturalness; MOD = moderate; SEV = severe, CB = cerebellar; SCA = spinocerebellar ataxia.

Duffy, Rosenbek, & Kent, 2007) participated in the study as experienced listeners. Clinically experienced listeners were selected over untrained listeners given their familiarity with dysarthria and attuned listening skills in an effort to increase the likelihood of higher intra- and interrater reliability (Zeplin & Kent, 1996). All listeners were native speakers of American English with no history of speech or language deficits per self-report. Listeners passed a bilateral pure-tone hearing screening at 25dB SPL, for the frequencies 500, 1000, 2000, and 4000 Hz and the vision screening described above. No listeners were excluded on the basis of these criteria. However, out of 11 persons originally recruited, one was unable to complete the study due to technical error. This yielded a final total of 10 experienced listeners—eight women and two men with a mean age of 43 years. The clinical practice setting of listeners varied across the continuum of care, with the majority being outpatient clinics. Years of clinical experience ranged from 5 to 40 years with a mean of 13.8 years ($SD = 10.8$).

Procedures

Speaker Session

Participants were seated at a table in a quiet room. Each participant wore an AKG C520 head-mounted microphone (constant mouth to microphone distance of 2 in.). The microphone was connected via an M-Audio preamplifier to the experimenter's computer running a custom written MATLAB speech recording program. The recording level was held constant for all speakers and the recordings were digitized at a sampling rate of 44000 Hz and quantization of 16 bits. To elicit the conversational speech sample, the recording software captured 3 min of the participant's conversational speech (elicited via predetermined, open-ended questions) without their awareness of being recorded. To do this, the experimenter pretended to "set up" equipment (e.g., placed the head-mounted microphone, manipulated knobs, etc.), in order to give the sense that the experiment had yet to start. Questions used to elicit conversation emerged organically from the conversation with the experimenter. Participants were asked after the recording of the covert conversation to determine their level of awareness of the recording. No participants suspected that they were being recorded. After the participant was made aware of the recording and reconsented in accordance with Institutional Review Board procedures, the experiment continued with the speech evaluation, consisting of sustained phonation, diadochokinetic rates, and the sentence portion of the speech intelligibility test for Windows (Yorkston, Beukelman, Hakel, & Dorsey, 2007) see Table 1 for a summary of intelligibility results). The participant was then offered a 10-min break while the examiner listened to the covert (conversation) speech recording and extracted the first eight sentences of five to 15 words in length that were free of proper nouns, low frequency words, formulaic expressions, and specialty vocabulary (see Sidtis et al., 2012, for detailed method)—five sentences to be used for analysis and three additional sentences to be used as a replacement if warranted. In this way, the experimenter was able to

create a set of stimuli that was the same for each individual speaker across the speaking conditions (covert conversation and overt reading). The sentences were typed into a Microsoft Word document using 20 pt. Arial font, double-spaced. Words were used if understandable given context. If the word was unintelligible, the experimenter verified the questionable word(s) with the speaker prior to the reading task. When the participant was ready, he or she was instructed to "read each sentence aloud using your typical speech" while being recorded. Participants read the sentences directly off the computer screen. The use of written text was considered an externally cued task as the text serves to denote word boundaries, and punctuation (e.g., commas) is known to influence prosody, such as breath pausing (see Huber et al., 2012; Steinhauer, 2003). The reading samples were recorded using the same procedure as the covert conversational recording, and the examiner ensured the mouth-to-microphone distance had not changed. The total experiment completion time for each participant ranged from 60–90 min.

To prepare files for the listener task, sentences from the covert conversation file were edited using sound editing software (Adobe Audition 9.0). Each participant had a final 10 sentences (5 covert conversation + the same 5 sentences read aloud), which were later used for rating by experienced listeners.

Listener Session

During the listener session, custom MATLAB software was used to present the recorded sentences in random order, as well as to obtain listener ratings. The total number of listening samples generated for the group with PD was: 10 speakers $\times$ 10 sentences (5 reading + 5 conversation) = 100 sentences plus 20 repeated files to test intrarater reliability (20% of the PD total), for a total of 120 sentences. For the group with CD, 6 speakers $\times$ 10 sentences (5 reading + 5 conversation) = 60 sentences plus 12 repeated files for reliability purposes (20% of the CD total), for a total of 72 sentences. Combining the PD and CD samples resulted in an overall total of 192 sentences. Each expert listener was then asked to make two judgments (understandability and naturalness) per sample resulting in 384 judgments per listener.

Following consent and the screening procedures, listeners were seated at a table with a computer monitor and wireless mouse. A 100-point visual analogue scale (VAS) was used for listener ratings; this method has previously been used to rate dysarthric speech (Spencer, Morgan, & Blond, 2009) and was recently found to be correlated with speech intelligibility (Stipancic, Tjaden, & Wilding, 2016). The VAS was displayed on the screen and could be used by the experimenter when providing task directions. Listeners were instructed to press the "PLAY" button when ready to start the listening session. By doing so, he or she would hear a sound file played only once. After listening to the entire sound file, the listeners were asked to use their best clinical judgment and rate each sample (i.e., a speaker's sentence in one of the two conditions) on two different dimensions—understandability and naturalness—by sliding

the cursor along the VAS. Throughout the session, the VAS for both dimensions was visible on the computer screen while each sample was played. The software program was designed to convert the position of the cursor on each VAS into a numerical value between 0 and 100 that was used for analysis. Perceptual dimensions and VAS end points were defined for listeners using the following definitions: understandability—"the speaker's articulatory precision," with the two ends of the scale labeled as "completely understandable" and "completely unable to understand"; and naturalness—"the speaker's prosody—defined as speech rate, rhythm, intonation, stress patterns and loudness" (Yorkston et al., 1999) with the two ends of the scale labeled as "completely natural" and "highly unnatural." Understandability is defined in numerous ways throughout the literature; the term has been used to represent general clarity of speech (Chepeha et al., 2016; Prathanee, Pumnum, Seepuaham, & Jaiyong, 2016; Seignemartin, Miranda, Luz, & Teixeira, 2015), to indicate clarity of an utterance in relation to prior context (Bloch, Saldert, & Ferm, 2015; Bloch & Wilkinson, 2004, 2009, 2011), and to describe incomplete contact of articulators (Commerford, 2000; Logemann & Fisher, 1981). In selecting understandability, it was our intent to have listeners focus on the broader notion of clarity of speech, while directing attention to nonprosodic aspects of speech in a brief/succinct way. Definitions of each dimension were provided on the computer monitor just above the VAS throughout the entire session. Task directions and definitions were provided in written form as well. Once the listener indicated full understanding of the task and definitions, they were asked to begin the rating session. Speech samples were presented through headphones (Sony Professional MDR-7506, Sony Corporation of America, New York, NY).

The MATLAB program was designed to track the sample number in a small window at the top of the screen. This allowed for the participant and experimenter to monitor progress. Each listener heard a random ordering of the original speech samples (100 PD + 60 CD = 160) first, followed by a random ordering of repeated speech samples (20% of total = 32) used to determine intrarater reliability. Headphone volume was set by an investigator with normal bilateral hearing thresholds (< 25 dB SPL, for the frequencies 500, 1000, 2000, and 4000 Hz). Once set, the volume remained at a constant level between listeners. The judges' ratings for all utterances produced by the individual participants were averaged by measure, yielding an overall score for each participant on understandability and naturalness for each speaking condition.

Data Analysis

Prior to analysis, listener ratings were collapsed across sentences, giving an average rating of understandability and naturalness for each speaker for each speaker condition. Between-groups comparisons were completed using two 2 (Etiology) $\times$ 2 (Speaking Task) analyses of variance (ANOVAs) and paired *t* tests for planned comparisons. Significance was assessed at $p < .025$ for all tests.

Results

Reliability

For reliability of the sentence extraction method, interrater reliability was calculated between the investigator and two research assistants who received training in the extraction procedure. Reliability was conducted on 38% of the total participants (six out of 16). A word-to-word comparison was used for each sentence, with contractions counting as one word. All measures of interrater reliability were calculated using the intraclass correlation coefficient (group averages model). A standard acceptance criteria of .7 was determined a priori (Nunnally & Bernstein, 1994). For the sentence extraction procedure, a reliability of .891 was demonstrated between raters.

To assess intrarater reliability, 20% of the samples were repeated for each listener and Cronbach's alpha was used. Intrarater reliability was sufficiently high for both understandability ($\alpha = .945$, range = .77–.97) and naturalness ($\alpha = .924$, range = .74–.94).

Interrater reliability was calculated to assess the level of agreement among the 10 listeners using the intraclass correlation coefficient (group averages model). A high degree of reliability was demonstrated for both understandability (.867, range = .83–.89) and naturalness (.874, range = .84–.89).

Group Analysis

Understandability

A 2 (Etiology) $\times$ 2 (Speaking Task) ANOVA revealed a significant main effect of task type on understandability, $F(1, 14) = 9.55$, $p = .008$, such that participants with both PD and CD were rated as significantly more understandable in the reading task ($M = 74.53$, $SD = 18.83$) than in the covert conversation task ($M = 62.98$, $SD = 24.49$). There was no significant main effect of etiology, $F(1, 14) = 0.002$, $p = .964$, and no interaction between etiology and task type, $F(1, 14) = 2.44$, $p = .141$. A paired *t* test comparing speaking tasks for participants with PD was conducted as a planned comparison, and revealed a significant difference, $t(1, 9) = 3.09$, $p = .013$, such that the reading task ($M = 76.29$, $SD = 22.19$) was rated as more understandable than the covert conversation task ($M = 60.84$, $SD = 29.03$). In a similar manner, a paired *t* test comparing task type for participants with CD revealed a significant difference, $t(1, 5) = 3.42$, $p = .019$, such that the reading task ($M = 71.61$, $SD = 12.72$) was rated as significantly more understandable than the conversation task ($M = 66.53$, $SD = 16.06$; see Figure 1).

Naturalness

A 2 (Etiology) $\times$ 2 (Task Type) ANOVA revealed a significant main effect of task type on naturalness, $F(1, 14) = 5.34$, $p = .037$, such that participants were rated as significantly more natural in the reading task ($M = 58.06$, $SD = 21.23$) than in the conversation task ($M = 50.69$, $SD = 21.31$). There was no significant main effect of etiology, $F(1, 14) =$

Figure 1. Paired *t* test of mean perceptual ratings of experienced listeners for understandability compared across speaking tasks for Parkinson's disease (PD) and cerebellar disease (CD). Error bars represent standard error for paired *t* tests.

0.002, $p = .964$. However, there was a significant interaction between etiology and speaking task, $F(1, 14) = 7.85$, $p = .014$. Participants with PD were perceived as speaking significantly more naturally in the reading condition ($M = 64.32$, $SD = 21.50$) than in the conversation condition ($M = 51.82$, $SD = 24.50$), $t(1, 9) = 3.38$, $p = .008$. However, participants with CD were not perceived to be significantly different in their naturalness for both reading ($M = 47.62$, $SD = 17.63$) and conversation conditions ($M = 48.17$, $SD = 16.61$), $t(1, 5) = 1.21$, $p = .279$ (see Figure 2).

Individual Participant Analyses

Given the heterogeneity inherent to these clinical populations, descriptive statistics of experienced listeners'

Figure 2. Paired *t* test of mean perceptual ratings of experienced listeners for naturalness compared across speaking tasks for Parkinson's disease (PD) and cerebellar disease (CD). Error bars represent standard error for paired *t* tests.

perceptual ratings for each participant were conducted. Results can be seen in Table 2. To evaluate individual performance patterns for each speaker, differences between speech tasks (conversation vs. reading) per perceptual dimension (naturalness and understandability) were determined for each speaker. A positive difference indicates that the speaker was rated as better during conversation than during reading, whereas a negative difference suggests that a speaker was rated as better during the externally cued reading task. Scores that represent a conservative difference of 15 points on the VAS (Bailey et al., 2012; Engen, 1971; Gallagher, Liebman, & Bijur, 2001; Kelly, 2001) were flagged as an indication of marked change in perceptual ratings between speech tasks.

On the basis of difference scores, four speakers with PD had a 15-point or more increase in understandability in the externally cued reading task compared to zero participants with CD. With respect to perceived naturalness, three speakers with PD had a 15-point or more increase in naturalness while reading compared to zero participants for the cerebellar group. Across perceptual categories, four speakers with PD showed a pronounced increase in one or both perceptual domains in the reading task; no speakers with CD showed a marked change, in either direction, during the reading task. The speaker with the largest difference across both understandability and naturalness was PD7. This participant was one of two speakers with PD rated as having severe hypokinetic dysarthria, and had the second-lowest intelligibility score at 82% ($M = 88.4$, $SD = 16.0$). Other factors, such as age, education, and years since PD diagnosis, were typical of group averages.

Discussion

The goal of the present study was to examine the effect of internally versus externally cued speech tasks on the understandability and naturalness of speakers with PD as rated by experienced listeners. A direct comparison was made between sentences extracted from a covertly recorded conversation (internally cued) and reading of the same sentences (externally cued) by the participants. Overall, the results of our investigation revealed that experienced listeners rated the speech of participants with PD as more natural and more understandable during the reading condition. The group with CD also demonstrated a difference between speaking conditions, but only for understandability; naturalness was not perceived to change between conversation and reading. Thus, the percept of naturalness appeared to be sensitive to capturing the differences between speakers with PD and CD.

Perceptual Ratings of Naturalness

Naturalness was defined for our experienced listeners as "the speaker's prosody—speech rate, rhythm, intonation, stress patterns, and loudness." On the basis of the listeners' ratings, speakers with PD showed a significant improvement in their naturalness from the conversation condition to the

Table 2. Mean (SD) perceptual ratings across speakers and tasks with difference scores.

Speaker	Understandability			Naturalness		
	Conversation	Reading	Difference ^a	Conversation	Reading	Difference
PD1	69.5 (19.9)	93.4 (10.0)	-23.9	52.5 (21.0)	80.7 (18.4)	-28.2
PD2	85.6 (14.3)	86.1 (13.8)	-0.5	75.3 (19.4)	75.9 (18.0)	-0.6
PD3	61.2 (24.6)	82.3 (14.2)	-21.1	50.5 (18.9)	65.0 (23.6)	-14.5
PD4	79.4 (16.7)	80.8 (16.2)	-1.4	55.9 (24.4)	61.6 (22.1)	-5.7
PD5	68.8 (22.4)	71.1 (24.4)	-2.3	52.1 (18.8)	50.6 (22.2)	1.5
PD6	4.2 (8.1)	19.1 (27.2)	-14.9	8.7 (11.1)	18.5 (18.7)	-9.8
PD7	15.7 (21.2)	69.1 (29.5)	-53.4	19.1 (18.2)	54.8 (27.0)	-35.7
PD8	77.1 (23.6)	90.5 (12.3)	-13.4	70.9 (24.6)	81.8 (18.8)	-10.9
PD9	91.7 (14.0)	96.9 (6.0)	-5.2	90.1 (12.0)	96.4 (9.9)	-6.3
PD10	55.2 (26.5)	73.6 (22.7)	-18.4	43.0 (23.0)	58.1 (22.5)	-15.1
<i>M</i>	60.8	76.3	-15.5	51.8	64.3	-12.5
<i>SD</i>	29.0	22.2	15.8	24.5	21.5	11.7
CD1	80.8 (18.7)	81.9 (14.4)	-1.1	63.7 (26.6)	60.1 (28.7)	3.6
CD2	39.7 (23.6)	49.4 (26.4)	-9.7	26.2 (19.8)	24.2 (22.0)	2.0
CD3	70.2 (23.2)	74.1 (19.3)	-3.9	57.3 (22.4)	59.6 (23.5)	-2.3
CD4	83.4 (19.9)	85.2 (21.6)	-1.8	68.1 (22.7)	68.6 (27.3)	-0.5
CD5	58.3 (24.9)	67.5 (22.7)	-9.2	37.2 (24.0)	33.2 (19.7)	4.0
CD6	66.8 (16.3)	71.6 (18.1)	-4.8	40.4 (23.4)	39.9 (21.8)	0.5
<i>M</i>	66.5	71.6	-5.1	48.8	47.6	-1.2
<i>SD</i>	16.1	12.7	3.6	16.6	17.6	2.4

Note. Rating scale ranged from 0–100 with 100 representing completely understandable or completely natural. PD = Parkinson's disease; CD = cerebellar disease.

^aDifference is covert conversation minus reading; bold values indicate those speakers whose difference is ≥ 15 points.

reading condition, whereas speakers with CD did not demonstrate this pattern. This finding has several possible explanations.

The most compelling and well-supported explanation is based on the underlying mechanisms that help individuals with PD perform motor skills more accurately when provided with external cues (Burleigh-Jacobs et al., 1997; Freeman, Cody, & Schady, 1993; Lehman et al., 2005; Majsak, 2000; Mak et al., 2013; McIntosh et al., 1997; Morris & Iansek, 1996; Thaut et al., 1996). There is a large body of limb motor control research demonstrating the benefit of external cues to motor performance in PD, such as a line on the floor to enhance gait, lines on the paper to improve micrographia, or a rhythmic auditory cue to facilitate thumb motion patterns (Burleigh-Jacobs et al., 1997; Chuma, Faruque Reza, Ikoma, & Mano, 2006; Cunningham et al., 1999; Oliveira et al., 1997). These cues purportedly assist basal ganglia function and/or bypass the disrupted mechanisms (Giladi et al., 2001; Morris, 2000). Without these cues, the impaired basal ganglia system relies on internal cueing mechanisms to scale movements accurately, which is often miscalibrated for individuals with PD. As a result, gait is often small and shuffling in nature, and writing is small and difficult to read. Although the internal cueing mechanisms of the basal ganglia are impaired in persons with PD, their ability to make use of external cues is still intact. As discussed earlier, printed text may not be considered a traditional external cue, but it may still act as an external trigger, leading our speakers with PD to favorably alter their prosodic aspects of speech.

An alternative explanation is that there are fewer demands on cognitive–linguistic functioning, specifically language formulation, when text is provided. As a result, the externally cued reading condition would presumably have fewer prosodic errors related to language formulation, such as disfluencies or filled pauses (Huber & Darling, 2011). In the internally cued conversational task, extra cognitive–linguistic processing is necessary to support language formulation and phoneme selection (van der Merwe, 1997). On the other hand, the reading task does not involve language formulation and subsequently has a reduced cognitive–linguistic load. Thus, when the message is predetermined by the provided text, frontolimbic circuitry is free to provide more resource support to the development and execution of motor programs mediated by the basal ganglia (Graybiel, Canales, & Capper-Loup, 2000; Turner & Desmurget, 2010). This argument was first put forth by Canter and Van Lancker (1985) and further substantiated by other investigators (Kempner & Van Lancker, 2002; Sidtis et al., 2012). Although a reduction in cognitive load is one explanation for more natural speech while reading, it is an unlikely explanation given that both groups would have been expected to improve in the externally cued condition.

Another possible reason for increased naturalness in the reading condition for speakers with PD may be increased attentional focus to the task of speaking. Externally cued tasks, by their very nature, often heighten a person's awareness and attention to the task. In fact, environmental factors such as use of speech materials and the physical setting

have been shown to increase typical speakers' attention to their speech production, enhancing performance (Goberman & Elmer, 2005; Hustad, 2007; Kent, Weismer, Kent, & Rosenbek, 1989). The same improvements have been found in individuals with PD (Keintz et al., 2007; Neely, 1956; Sarno, 1968; Weismer, 1984). Thus, it is possible that the participants with PD had heightened attention during the reading task versus the covertly recorded conversation, and this enhanced attention may have led to more natural-sounding speech. However, both speaker groups would have been expected to improve in the reading condition if the source of the difference was attentional focus.

Unlike participants with PD, the cerebellar group did not differ in their perceived naturalness between reading and covert conversation. The cerebellum is believed to play a prominent role in the generation of internal models for movement (Kawato, 1999), including speech (Duffy, 2013), and in the response to externally cued movements (Jueptner & Weiller, 1998; van Donkelaar & Staub, 2000). Although both the basal ganglia and cerebellum are concerned with motor performance, the cerebellum is preferentially involved in monitoring the outcome of movements by comparing them with sensory inputs (Jueptner & Weiller, 1998). When impaired, it is less likely that the cerebellum would be able to utilize external cues to adjust a movement (Iosa, Morone, Fusco, & Paolucci, 2013). In the present study, it is possible that the speakers with ataxia were unable to make the adjustments needed to improve the subtle nuances involved in the prosodic features of connected speech, whereas for those with PD, the ability to benefit from external cueing and incoming sensory stimuli is still intact, allowing for prosodic alteration. Another possibility may simply be due to reduced ability to improve naturalness compared with participants with PD.

Perceptual Ratings of Understandability

In the current study, understandability was defined for our experienced listeners as "the speaker's articulatory precision," which they used to rate each speech sample along a VAS as "completely understandable" to "completely unable to understand." Our attempt to focus listeners on articulation as the primary contributor to understandability, although less conventional, served as a distinct counterpart to the focus on naturalness/prosody. Results revealed a significant improvement in understandability for the reading task compared to the covert conversation for *both* speaker groups, which was contrary to our prediction. Ratings of understandability may have been influenced by the reduced cognitive-linguistic load of the reading task, and/or the increased attentional focus typical of a reading task.

As discussed earlier, the need for less cognitive-linguistic processing load may have helped to redirect more cognitive resources to the task of speaking. Bunton and Keintz (2008) demonstrated this phenomenon in their single-versus dual-task paradigm.

Intelligibility scores for reading significantly improved from 78.28% in the dual-task condition to 90.2% in the

single-task condition for participants with PD, which the authors attributed to a reduction in cognitive load. It is possible that the reading task in the current experiment allowed for more cognitive resources to be allocated to the mechanics of speech production, regardless of the speaker group. In the conversational speech task, additional higher-up processing had to occur prior to speech production, placing a greater demand on resource allocation (Bohland, Bullock, & Guenther, 2010; Tourville & Guenther, 2011; Van der Merwe, 1997). Thus, it is feasible that the result of a lessened cognitive load would lead to improved understandability during the reading task for both impairment groups (Kempler & Van Lancker, 2002).

Another possibility for an enhancement in perceived understandability in both groups may lie in the increase of attentional focus during performance of the reading task. As discussed earlier, the presence of recording and other testing equipment may change the execution of the task, inadvertently improving performance in production of individual phonemes, and/or increased amplitude of the speech signal (Aronson, 1990). As a result, the mechanics of speech productions were such that a noticeable improvement was perceived during the reading task for both speaker groups (Hustad & Weismer, 2007). However, increased attentional focus would have been expected to translate to improvement across perceptual ratings of naturalness and understandability for both groups.

Individual Performance Patterns

There were four speakers who had a pronounced (> 15 points) difference between the internally and externally cued conditions: PD1, PD3, PD7, and PD10. Of these speakers, PD7 had an exceptionally large increase in both understandability and naturalness during the reading condition. The individual performance patterns suggest that there is a subgroup of speakers with PD who experience a dramatic improvement to speech during reading. No participants from the group with CD had differences > 9.7, and the overall difference scores were quite small (mean differences between conversation and reading were 5.1 for understandability and 1.2 for naturalness, versus for the group with PD, which was 15.5 points difference for understandability and 12.5 for naturalness). Hence, additional research is needed to identify the specific speech parameters that may be linked to the listener judgments. A follow-up study examining rate of speech, dysfluencies, and acoustic indices is underway (Ballestracci, 2015).

Clinical Implications

Converging evidence suggests that clinicians should be mindful of the impact that external cues have on speech production and how their use during testing can paint an unrealistic picture of true day-to-day conversational ability. This study highlights the need for our testing to focus on representative samples and suggests that our elicitation instructions can influence speech performance. Clinicians

historically have placed much emphasis on the importance of intelligibility levels to diagnose and manage dysarthria. However, relying solely on intelligibility testing that uses reading (or repeating) stimuli may be limiting, and may inflate the perception of patients' true abilities, as indicated by results of the current study. Findings from the current investigation highlight the importance of using naturalness and, if possible, a covert/natural conversational speech sample in the assessment process.

Conclusions and Limitations

Speakers with PD appeared to experience benefits to both perceived understandability as well as perceived naturalness in an externally cued reading condition, whereas those with CD only increased their understandability during the reading task. Thus, the group with PD showed distinct benefits to prosody during the externally cued reading condition.

The main limitation of the current investigation is the small and unequal sample size. As is the case with many studies of clinical populations, the small number of participants can bring into question the strength and generalizability of the results. In addition, the potential effects of varied linguistic contexts of utterances across speakers warrant consideration. Although content was controlled between cueing conditions, variations in sentence structure and linguistic context between speakers may have affected the reading performance and, subsequently, listener ratings (Huber et al., 2012; Pell et al., 2006).

Future research is needed to better understand what constitutes an external speech cue, to examine how the use of external cueing influences speech production in participants with PD, and to further differentiate the effects of cueing from related issues, such as the degree of structure of the speaking task (Huber & Darling, 2011). Acoustic measures of speech production comparing the underlying features of understandability and naturalness in conversational speech, reading, and/or repeating would lend additional insights to the growing body of literature exploring internal versus external cueing differences in PD. Continued exploration into the similarities and differences between speech and limb motor control regarding internal and external cueing mechanisms would add to our understanding of what constitutes an external cue for speech tasks. It would also be advantageous to identify ways to standardize the rating of speech characteristics, particularly those aligned with prosody, during a spontaneous speech sample. Improvements in these areas will help to enhance understanding of external cueing in PD and CD, allowing clinicians to provide better service and education to their patients and families.

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