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Language abilities of patients with primary progressive multiple sclerosis: A preliminary group and case investigation

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Abstract

Language impairments are reported in multiple sclerosis (MS). To date, the majority of studies have evaluated language differences between relapsing-remitting (RR) and chronic progressive (CP) clinical courses. Neurologists have distinguished two progressive courses of MS: primary progressive (PP) MS and secondary progressive (SP) MS. Recent evidence suggests that cognitive performance profiles may provide a means of differentiating between the clinical courses of RR, SP, and PPMS. With this in mind, a deviation of language profiles between sub-types is predicted. The purpose of this study is to profile the language abilities of five participants with PPMS. Five participants with PPMS participated in this investigation. The participants were assessed using the Neurosensory Center Comprehensive Examination for Aphasia (NCCEA), the Boston Naming Test (BNT), and the Test of Language Competence–Expanded (TLC-E). Data analysis consisted of (a) comparison of the total scores achieved by the PPMS participants and a group of 26 age-matched controls on the NCCEA, BNT, and TLC-E, and (b) case studies to individually profile the language abilities of the five participants with PPMS. Comparison of the NCCEA, BNT, and TLC-E total scores of the participants with PPMS and the control group did not indicate significant differences between the two groups. Case-by-case analysis revealed deficits in meta-linguistic abilities in two participants. The results provide preliminary evidence to suggest that, although patients with PPMS may have preserved general language abilities, some individuals may present with mild impairments in high-level linguistic abilities.

Keywords: Language, high-level language, multiple sclerosis, primary progressive multiple sclerosis, MS, PPMS.

Introduction

Multiple sclerosis (MS) is an acquired primary demyelinating disease which affects the central nervous system, triggering a variety of identifiable physical and intellectual clinical characteristics (Brassington & Marsh, 1998; Whitaker & Mitchell, 1997). The clinical course of MS typically follows one of three patterns (Lublin & Reingold, 1996). Relapsing-remitting (RR) MS comprises of relapses with full recovery or a sequelae and residual deficit upon recovery. The periods between relapses are indicated by lack of disease progression or episodic acute periods of worsening interspersed with periods of stability or improvement. A lesser known clinical course, termed "Benign MS", is also described and is characterized by very occasional relapses, which are mild in nature with a good recovery. Primary progressive (PP) MS is distinguished by gradual, almost continuous deterioration in function with minor fluctuations but no distinct relapses. Secondary progressive (SP) MS is characterized by an initial relapsing-remitting course followed by progression, with or without occasional relapses, minor remissions, and plateaux. SPMS is the natural evolution of RRMS, with ~90% of individuals with RRMS having SPMS after a 26-year follow-up (Weinshenker, Bass, Rice, Noseworthy, Carriere, Baskerville, et al., 1989). PPMS is the least common course of MS. Approximately 15% of MS patients are initially diagnosed with PPMS, with nearly a third eventually experiencing one or more relapses (Wolinsky, 2003). Whether RRMS, PPMS, and SPMS differ in prognosis, epidemiology, pathology, Magnetic Resonance Imaging findings, and cognitive abilities remains undetermined (Kraus, Schutze, Brokate, Kroger, Schwendemann, & Hildebrandt, 2005; Palace, 2003; Thompson, Polman, Miller, McDonald, Brochet, Filippi, et al., 1997; Vukusic & Confavreux, 2003; Wolinsky, 2003).

Cognitive deficits are reported to be most severe in SPMS, followed by PPMS, then RRMS (Comi, Filippi, Martinelli, Campi, Rodegher, Alberoni, et al., 1995; DeSonneville, Boringa, Reuling, Lazeron, Adèr, & Polman, 2002; Gaudino, Chiaravalloti,

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DeLuca, & Diamond, 2001; Huijbregts, Kalkers, de Sonneville, de Groot, Reuling, & Polman, 2004). Nonetheless, the literature in its entirety does not support this premise. For example, Foong, Rozewicz, Chong, Thompson, Miller, and Ron (2000) showed that PPMS and SPMS patients could not be differentiated based on cognitive performance. These authors found a negligible difference in the proportion of PPMS and SPMS patients who had severe cognitive impairment. In addition, Wachowius, Talley, Silver, Heinze, and Sailer (2005) found that PPMS patients performed worse than SPMS patients on verbal fluency and verbal learning tasks.

Preceding 1996, neurologists did not clinically distinguish PPMS and SPMS and as such classified both courses as chronic progressive (CP) MS (Bennett, Ditmar, & Raubach, 1991). Researchers examining cognitive dysfunction in MS now distinguish between PPMS, SPMS, and RRMS. To date, research examining language processing in MS has utilized the terminology from the older classification system (i.e., CP and RR MS), thus indicating a deficient knowledge of language abilities in the sub-types of progressive MS.

Language impairments often co-occur with cognitive deficits in both CPMS and RRMS sub-types of MS (Friend, Rabin, Groninger, Deluty, Bever, & Grattan, 1999; Lethlean & Murdoch, 1997). Lethlean and Murdoch (1997) described a general language profile of 60 patients with stable MS. These authors reported that their cohort of MS patients had difficulty with vocabulary, semantic tasks, understanding ambiguous sentences and metaphoric expressions, making inferences, and re-creating sentences. Other studies have reported deficits in phonemic fluency (Beatty, 2002; Beatty, Goodkin, Beatty, & Monson, 1989; Beatty, Hames, Blanco, Paul, & Wilbanks, 1995; Friend et al., 1999; Lethlean & Murdoch, 1993), semantic fluency (Beatty, 2002; Beatty et al., 1989, 1995; Friend et al., 1999), picture naming (Friend et al., 1999; Lethlean & Murdoch, 1994; Zakzanis, 2000), auditory comprehension (Friend et al., 1999; Laatu, Hamalainen, Revonsuo, Portin, & Ruutiainen, 1999; Zakzanis, 2000), and reading comprehension (Grossman, Robinson, Onishi, Thompson, Cohen, & D'Esposito, 1995). Based on the findings of published investigations to date, the language profiles of CPMS and RRMS is believed to be comparable, however evidence suggests that individuals with CPMS may have a greater degree of language dysfunction than individuals with RRMS (Friend et al., 1999; Lethlean & Murdoch, 1994; Zakzanis, 2000).

The high prevalence of language deficits in people with CPMS suggests that patients with PPMS and SPMS have a pre-disposition to language dysfunction. In the fact that the progressive phase of MS is associated with irreversible accumulation of disability, both physical and intellectual in nature (Vukusic & Confavreux, 2003), PPMS and SPMS patients may have more severe language difficulties than their RRMS counterparts. The language abilities of PPMS patients are of particular interest, as reports concerning the cognitive abilities in this form of MS are largely contradictory. Comi et al. (1995) reported that only 7% of participants with PPMS in their study had cognitive deficits. In contrast, Wachowius et al. (2005) found that 37% of participants with PPMS studied had cognitive deficits. Preliminary findings concerning language abilities of PPMS patients have shown normal performance on the Token Test and phonemic fluency tasks (Comi et al., 1995). These outcomes are divergent from a study by Wachowius et al. (2005), who reported impaired performance on a phonemic fluency task in participants with PPMS. Similarly, Huijbregts et al. (2004) reported impaired semantic fluency in a cohort of PPMS participants. It is clear that the extent to which language abilities are reduced in PPMS remains elusive.

The purpose of the current study is to comprehensively profile the language abilities of five participants with PPMS. First, the language results of the study participants will be compared to those of a neurologically healthy control group. Second, the individual language profiles of these five participants will be presented and examined. Given a high degree of performance variability within the MS literature (Beatty et al., 1995; Grossman et al., 1995; Laatu et al., 1999; Lethlean & Murdoch, 1997), this approach will allow for the detection of individual language impairments that may otherwise be concealed with the implementation of group-wise comparisons.

Method

Participants

Five participants with PPMS, diagnosed by a neurologist following a comprehensive neurological examination, including a MRI scan, participated in the present study. PPMS diagnosis was based on criteria outlined in a position paper by Thompson, Montalban, Barkhof, Brochet, Filippi, Miller, et al. (2000). The participants were recruited via major Brisbane metropolitan hospitals and the MS Society of Queensland. Exclusion criteria included: a history of neurological disease or disorder in addition to MS; a speech and/or language disorder prior to diagnosis of MS; a history of drug and/or alcohol abuse; and/or English as a second language. All participants had intelligible speech as judged by a qualified speech-language pathologist. Clinical and demographic information pertaining to the five participants with PPMS is shown in Table I. Four female participants and one male participant participated in the study. PPMS participants were aged between 55–77 years (mean age = 63.1 years, SD = 10 years) with a level of education from 8-17 years (mean

| Clinical/domographia | Participant | | | | | | | | | | |
|----------------------------------|---|---|---|---|------------------|--|--|--|--|--|--|
| Clinical/demographic information | 1 | 2 | 3 | 4 | 5 | | | | | | |
| Age | 55 | 70 | 56 | 77 | 57 | | | | | | |
| Gender | Female | Female | Female | Male | Female | | | | | | |
| Education (years) | 15 | 17 | 12 | 16 | 8 | | | | | | |
| Motor & sensation | Independent | Wheelchair; reduced sensation in left hand | Wheelchair | Wheelchair; limited upper limb mobility and sensation | Wheelchair | | | | | | |
| Vision & hearing | Corrected vision; reduced acuity in right ear | Corrected vision; history of optic neuritis; reduced acuity | Corrected vision; history of optic neuritis | Corrected vision | Corrected vision | | | | | | |
| Disease duration | 2 years | 36 years | 24 years | 23 years | 4 years | | | | | | |
| Medication | Betaferon | Ditropan | Endep | Micardis | Temaze | | | | | | |
| | Anti-depressants | Endep | - | Baclofen | | | | | | | |
| | | Valium | | | | | | | | | |
| | | Normison | | | | | | | | | |

Table I. Clinical and demographic information for participants with primary progressive multiple sclerosis.

Betaferon = immuno-modifier; Ditropan = treats bladder dysfunction; Endep = anti-depressant; Valium = anti-anxiety agent; Normison = sedative; Micardis = anti-hypertensive.

education = 13.8 years, SD = 3.6 years). Disease duration ranged from 2–36 years (mean disease duration = 17.8 years, SD = 14.5 years).

Twenty-six age-matched, English speaking individuals (14 males, 12 females) (mean age = 60.15 years, SD = 14.04 years; mean level of education = 14.04 years, SD = 4.1 years) served as a control group. The control participants had no reported history of neurological impairment, substance abuse, psychiatric disorder, or speech and/or language disorder. All control participants performed within the intact range on either the Dementia Rating Scale or the Dementia Rating Scale-2 (Mattis, 1988).

The Dementia Rating Scale-2 (DRS-2) (Mattis, 1988) was used to assess the cognitive functioning of each PPMS participant (refer to Table II). The Total Scores for three participants (Participants 2, 3, and 5) fell within the intact range, with Age and Education Corrected Mayo Older Americans Normative Studies Scaled Scores (AEMSS) of 10, 11, and 11, respectively. One participant (Participant 1) achieved an AEMSS of 5, and this score fell within the moderately impaired range. One participant (Participant 4) achieved an AEMSS of 6, and this score fell within the mildly impaired range.

Procedure

The five participants with PPMS were administered a concise, yet comprehensive, battery of language assessments. The employed assessments were selected as they have demonstrated sensitivity to changes in general and high-level language performance in other populations (e.g., traumatic brain injury). Specifically the battery included:

- 1. The Neurosensory Center Comprehensive Examination for Aphasia (NCCEA) (Spreen & Benton, 1969) is an assessment of general language abilities which evaluates picture and tactile naming, repetition, sentence construction, word fluency, auditory comprehension, reading comprehension, writing, and articulation.
- The Boston Naming Test (BNT) (Kaplan, Goodglass, & Weintraub, 1983) tests the ability to label black and white line drawings of objects which vary in word frequency.
- 3. The Test of Language Competence-Expanded (TLC-E) (Wiig, & Secord, 1989) assesses meta-linguistic competence in semantics, syntax, and/or pragmatics through interpreting ambiguous sentences, making inferences,

| | Participant 1 | | Participant 2 | | Participant 3 | | Participant 4 | | Participant 5 | |
|-----------------------------------|---------------|---------|---------------|---------|---------------|---------|---------------|---------|---------------|---------|
| DRS-2 scale | Raw score | % range |
| Attention (37) | 36 | 60-71 | 37 | 82–89 | 36 | 60-71 | 37 | 82-89 | 36 | 60-71 |
| Initiation/ perseveration (37) | 27 | 2 | 37 | 60–71 | 37 | 60-71 | 26 | 1 | 37 | 60–71 |
| Construction (6) | 6 | 41-59 | 6 | 41-59 | 6 | 41-59 | 5 | 11–18 | 6 | 41-59 |
| Conceptualization (39) | 35 | 19–28 | 36 | 41-59 | 39 | 72-81 | 39 | 82-89 | 35 | 19–28 |
| Memory (25) | 24 | 41-59 | 24 | 41-59 | 22 | 11–18 | 23 | 41-59 | 24 | 41-59 |
| DRS-2 Total Score (144) | 128 | 6–10 | 140 | 60–71 | 140 | 60-71 | 130 | 11–18 | 138 | 41–59 |

Figures in italics represent reduced performance.

recreating sentences, and interpreting figurative language. The TLC-E also has a supplemental memory test (remembering word pairs).

The assessments were conducted in a quiet distraction-free environment according to the instructions provided in each test manual.

Data analysis

Data analysis consisted of (a) comparison of the total scores achieved by the participants with PPMS and the control group on the NCCEA, BNT, and TLC-E, and (b) case studies to profile the individual language abilities of the five persons with PPMS. Mann-Whitney U-tests were used to compare the *total mean scores* achieved by the PPMS group and the control group on the NCCEA, BNT, and TLC-E. The Mann-Whitney U-test was employed because the samples used in the current study were unequal in size.

Case studies were used to further describe the language data collected from each participant. Total scores for each participant on the NCCEA, BNT, and TLC-R were reported. The scores achieved by each participant on the 20 sub-tests of the NCCEA and five sub-tests of the TLC-E were also reported. A conservative criterion for impaired performance of ≥ 2 SD below the mean of the control group on each test or sub-test was used. In instances where the control group had a SD of 0, any score below the mean was considered in the impaired range.

Results

Comparison of NCCEA, BNT, and TLC-E total scores

The mean and SD for the PPMS group and control group for NCCEA total score, BNT total score, and TLC-E total score are presented in Table III. A series of Mann-Whitney U-tests demonstrated that there were no significant differences between the groups on the total score of the NCCEA (U = 52.5, p = .912), BNT (U = 61.0, p = .828), or the TLC-E (U = 52.5, p = .514).

Table III. Neurosensory Centre Comprehensive Examination for Aphasia (NCCEA), Boston Naming Test (BNT), and Test of Language Competence–Expanded (TLC-E) total scores, means, and standard deviations (SD) for the primary progressive multiple sclerosis participant group and the control group.

| | - | MS ipants = 5) | Control (n= | 0 1 | – Mann-Whitney |
|-----------------------|------------------------|------------------------|---------------------------|------|--|
| Test | Mean | lean SD Mean | | | U-tests |
| NCCEA BNT TLC-E | 570.9 56.6 173.2 | 21.58 2.30 18.90 | 562.13 55.77 179.92 | 3.84 | U = 52.5, p = .912 $U = 61.0, p = .828$ $U = 52.5, p = .514$ |

Case descriptions

Participant 1. Participant 1 was a 55-year-old woman with 15 years of education, who was diagnosed with MS 2 years prior to entering the study. Her DRS-2 score indicated a moderate cognitive impairment. According to the DRS-2, attention, construction, and memory abilities were within normal limits but performance was impaired on initiation/perseveration and conceptualization sub-tests (see Table II). An NCCEA total score of 551 was within 2 SD of the control group mean. Scores on all but two sub-tests of the NCCEA were within 2 SD of the control group. Scores on Writing, copying, and Articulation sub-tests were within the impaired range (see Table IV). Participant 1's BNT total score of 57 was within 2 SD of the control group mean. A TLC-E total score of 170 was within 2 SD of the control group mean, as were scores on all TLC-E sub-tests except for Making inferences. The score achieved on the Making inferences sub-test was in the impaired range (see Table V).

Participant 2. Participant 2 was a 70-year-old woman with 17 years of education, who was diagnosed with MS 36 years prior to entering the study. Her DRS-2 score indicated intact cognition. An NCCEA total score of 588 was within 2 SD of the control group mean. Scores on all but one sub-test of the NCCEA were within 2 SD of the control group mean. The score achieved on *Tactile naming, left hand* sub-test was in the impaired range (see Table IV). Participant 2's BNT total score of 57 was within 2 SD of the control group mean. The score of 57 was within 2 SD of the control group mean as were scores on the TLC-E sub-tests (see Table V).

Participant 3. Participant 3 was a 56-year-old woman with 12 years of education, who was diagnosed with MS 24 years prior to entering the study. Her DRS-2 score indicated intact cognition, although her performance was impaired on the memory sub-tests (see Table II). An NCCEA total score of 554 was within 2 SD of the control group mean. Scores on all but one sub-test of the NCCEA were within 2 SD of the control group. The score achieved on the *Articulation* sub-test was in the impaired range (see Table IV). Participant 3's BNT total score of 59 was within 2 SD of the control group mean. Her TLC-E total score of 164 was within 2 SD of the control group mean, as were her scores on the TLC-E sub-tests (see Table V).

Participant 4. Participant 4 was a 77-year-old man with 16 years of education, who was diagnosed with MS 23 years prior to entering the study. His DRS-2 score indicated a mild cognitive impairment. According to the DRS-2, attention, conceptualization, and memory abilities were within normal limits, but

Table IV. Neurosensory Centre Comprehensive Examination of Aphasia (NCCEA) results for participants with primary progressive multiple sclerosis and the control group.

| | Participant 1 | | Participant 2 | | Partic | Participant 3 | | Participant 4 | | Participant 5 | | Controls $(n=26)$ | |
|---------------------------------------|---------------|-------------------------|---------------|-------------------------|--------|-------------------------|-------|---------------|-------|-------------------------|--------|-------------------|--|
| NCCEA sub-test | Score | $\uparrow\downarrow$ SD | Score | $\uparrow\downarrow$ SD | Score | $\uparrow\downarrow$ SD | Score | ↑↓ SD | Score | $\uparrow\downarrow$ SD | Mean | SD | |
| Visual naming (28) | 16 | 1.20 | 16 | 1.20 | 16 | 1.20 | 16 | ↑.20 | 16 | 1.20 | 15.96 | .20 | |
| Description of use (16) | 16 | .00 | 16 | .00 | 16 | .00 | 15 | \downarrow | 16 | .00 | 16.00 | .00 | |
| Tactile naming, right hand (16) | 16 | ↑.20 | 16 | ↑.20 | 16 | 1.20 | 0 | ↓79.8 | 16 | 1.20 | 15.96 | .20 | |
| Tactile naming, left hand (16) | 16 | 1.20 | 15 | <i>↓4</i> .8 | 16 | 1.20 | 2 | ↓69.8 | 15 | <i>↓4.8</i> | 15.96 | .20 | |
| Sentence repetition (27) | 18 | 1.43 | 25 | 12.97 | 16 | ↓.29 | 21 | 1.51 | 17 | 1.07 | 16.81 | 2.76 | |
| Repetition of digits (18) | 9 | ↓.51 | 9 | ↓.51 | 9 | ↓.51 | 9 | ↓.51 | 10 | 1.02 | 9.96 | 1.89 | |
| Reversal of digits (18) | 6 | ↓1.21 | 8 | 1.13 | 8 | 1.13 | 7 | ↓.54 | 10 | 1.46 | 7.81 | 1.50 | |
| Word fluency | 36 | $\downarrow.64$ | 58 | 1.90 | 37 | ↓.57 | 47 | 1.13 | 25 | \downarrow 1.41 | 45.15 | 14.34 | |
| Sentence construction (25) | 25 | 1.30 | 25 | 1.30 | 25 | 1.30 | 25 | 1.30 | 25 | 1.30 | 24.69 | 1.05 | |
| Identification by name (16) | 16 | .00 | 16 | .00 | 16 | .00 | 16 | .00 | 16 | .00 | 16.00 | .00 | |
| Token test (163) | 162 | 1.32 | 162 | 1.32 | 161 | 1.02 | 162 | 1.32 | 159 | ↓.56 | 160.92 | 3.42 | |
| Oral reading (names) (20) | 20 | 1.19 | 20 | ↑.19 | 20 | 1.19 | 19 | ↓.32 | 20 | ↑.19 | 19.62 | 1.96 | |
| Oral reading (sentences) (16) | 16 | .00 | 16 | .00 | 16 | .00 | 16 | .00 | 16 | .00 | 16.00 | .00 | |
| Reading names for meaning (10) | 10 | .00 | 10 | .00 | 10 | .00 | 16 | .00 | 10 | .00 | 10.00 | .00 | |
| Reading sentences for meaning (17) | 17 | .00 | 17 | .00 | 17 | .00 | 17 | .00 | 17 | .00 | 17.00 | .00 | |
| Visual-graphic naming (8) | 8 | 1.36 | 8 | 1.36 | 8 | 1.36 | 0 | <i>↓23.88</i> | 8 | 1.36 | 7.88 | .33 | |
| Writing of names (24) | 24 | 1.39 | 24 | 1.39 | 24 | 1.39 | 0 | ↓24.10 | 24 | 1.39 | 23.62 | .98 | |
| Writing to dictation (14) | 14 | 1.69 | 14 | ↑.69 | 14 | 1.69 | 0 | ↓5.85 | 14 | 1.69 | 12.52 | 2.14 | |
| Writing (copying) (12) | 10 | <i>↓2.82</i> | 12 | 1.62 | 12 | 1.62 | 0 | ↓25.04 | 12 | 1.62 | 11.27 | .45 | |
| Articulation (102) | 96 | ↓5.82 | 101 | 1.93 | 97 | ↓4.47 | 83 | <i>↓23.39</i> | 102 | 12.28 | 100.31 | .74 | |
| Total score | 551 | ↓.58 | 588 | 1.34 | 554 | $\downarrow.42$ | 465 | ↓5.05 | 548 | $\downarrow.74$ | 562.13 | 19.22 | |

Figures in italics represent reduced performance.

performance was impaired on the initiation/ perseveration and construction sub-tests (see Table II). An NCCEA total score of 465 was 5.05 SD below the control group mean and was in the impaired range. Scores on *Description of use, Tactile naming, right hand, Tactile naming, left hand; Visualgraphic naming; Writing of names; Writing to dictation; Writing, copying;* and *Articulation* were also in the impaired range. Scores on the remaining sub-tests of the NCCEA were within 2 SD of the control group mean (see Table IV). Participant 4's BNT total score of 54 was within 2 SD of the control group mean. The TLC-E total score of 159 was within 2 SD of the control group mean as were his scores on the TLC-E sub-tests (see Table V).

Participant 5. Participant 5 was a 57-year-old woman with 8 years of education, who was diagnosed with MS 4 years prior to entering the study. The DRS-2 indicated intact cognition, although performance

was impaired on the conceptualization sub-test (see Table II). An NCCEA total score of 548 was within 2 SD of the control group mean. Scores on all but one sub-test of the NCCEA were within 2 SD of the control group mean. The score achieved on the *Tactile naming, left hand* sub-test was in the impaired range (see Table IV). Participant 5's BNT total score of 53 was within 2 SD of the control group mean. A TLC-E total dcore of 143 was 2.97 SD below the control group mean and was in the impaired range. Scores on the *Making inferences* and *Figurative language* sub-tests were in the impaired range. Scores on the remaining three sub-tests of the TLC-E were within 2 SD of the control group mean (see Table V).

Discussion

To our knowledge, this is the first study to examine in detail the language abilities of participants with

| | Participant 1 | | Participant 1 Participant 2 1 | | | Participant 3 Par | | | Participant 4 | | Participant 5 | | rols 26) |
|-----------------------------|---------------|-------------------|-------------------------------|------------------|-------|-------------------|-------|-------------------|---------------|-------------------|---------------|-------|-------------|
| TLC-E sub-test | Score | ↑↓sd | Score | ↑↓sd | Score | ↑↓sd | Score | ↑↓sd | Score | ↑↓sd | Mean | SD | |
| Ambiguous sentences (39) | 37 | 1.39 | 33 | ↓.28 | 34 | ↓.11 | 33 | ↓.28 | 27 | ↓1.28 | 34.65 | 5.97 | |
| Making inferences (36) | 25 | $\downarrow 2.08$ | 30 | $\downarrow.49$ | 28 | ↓1.12 | 28 | ↓1.12 | 20 | ↓3.66 | 31.54 | 3.15 | |
| Recreating sentences (78) | 72 | \downarrow .26 | 70 | ↓.77 | 68 | ↓1.28 | 66 | \downarrow 1.79 | 68 | ↓1.28 | 73.04 | 3.94 | |
| Figurative language (36) | 36 | 1.92 | 36 | 1.92 | 34 | $\uparrow.04$ | 32 | ↓.85 | 28 | \downarrow 2.62 | 33.92 | 2.26 | |
| Remembering word pairs (32) | 10 | ↓.39 | 5 | \downarrow .87 | 7 | $\downarrow.74$ | 1 | ↓1.43 | 14 | 1.07 | 13.38 | 8.66 | |
| Total score (221) | 170 | ↓.80 | 169 | ↓.88 | 164 | ↓1.28 | 159 | ↓1.68 | 143 | ↓2.97 | 179.92 | 12.42 | |

Table V. Test of Language Competence-Expanded Edition (TLC-E) results for participants with primary progressive multiple sclerosis and the control group.

Figures in italics represent reduced performance.

PPMS, particularly on more complex high-level language structures. Comparison of the NCCEA, BNT, and TLC-E total scores of the participants with PPMS and the control group did not indicate any significant differences between the two groups. In contrast, case-by-case analysis revealed impaired performance on selected language sub-tests in all five participants with PPMS.

Although each participant with PPMS presented with impaired performance on some of the language tests used, not all of these deficits could be attributed to language dysfunction. Rather, the deficits demonstrated by participants on most sub-tests of the NCCEA (Tactile naming, left hand; Tactile naming, right hand; Writing, copying; Visual-graphic naming; Writing of names; Writing to dictation; and Articulation) were more likely a consequence of sensory and/or motor problems associated with MS disease progression than impaired language processing. In accordance with these present observations, other studies have discussed the impact of sensory and motor problems found in participants with MS on NCCEA test performance (FitzGerald, Murdoch, & Chenery, 1987; Lethlean & Murdoch, 1993).

When NCCEA sub-tests influenced by sensory and motor abilities were excluded, two participants (Participants 2 and 3) scored within 2 SD of the control group mean on all language tests and three participants had deficits on selected sub-tests of the test battery. One participant (Participant 4) scored within the impaired range for NCCEA Description of use. Participant 4's impaired score on NCCEA Description of use may not be theoretically or clinically important as his score of 15 out of a total of 16 was only one point below the control group mean score of 16 (SD = 0). Of greater interest are the two participants who presented with impaired performance on TLC-E. Lethlean and Murdoch (1997) have previously demonstrated that MS patients have difficulties performing the tasks of the TLC-E, an examination of meta-linguistic competence. Participant 1 scored within the impaired range on TLC-E Making inferences sub-test and Participant 5 scored within the impaired range on TLC-E Making inferences and Figurative language sub-tests, and total score. However, it is probable that Participant 5's

scores on the TLC-E may have been influenced, at least in part, by her level of education (8 years).

In this study, impairment was defined as a score less than or equal to 2 SD below the mean of the control group sample on a given test or sub-test. Recent studies have set less conservative cut-offs for impairment, at less than or equal to 1 SD (Heaton, Taylor, & Manly, 2003) and less than or equal to 1.5 SD (Cook, Murdoch, Cahill, & Whelan, 2004) below the mean of the control group. Use of 1 SD below the mean as the criterion for abnormal performance would have influenced interpretation of the TLC-E results in the current study. Four of the five participants with PPMS would have fallen within the impaired range for one or more sub-tests of this assessment. Specifically, three participants would have been impaired on the TLC-E total score (Participants 3, 4, and 5), one participant on Associations (Participant 5), three participants on Recreating sentences (Participants 3, 4, and 5), one participant on Figurative language (Participant 5), and one participant on Remembering word lists (Participant 4). In addition, one participant (Participant 5) would have had impaired phonemic fluency (NCCEA).

This study provides emerging evidence to suggest that individuals with PPMS may have preserved general language abilities with potential mild deficits in meta-linguistic abilities in some individuals. Deficits in picture naming, vocabulary, phonemic fluency, and auditory comprehension previously reported in individuals with CPMS (Friend et al., 1999; Grossman et al., 1995; Lethlean & Murdoch, 1993, 1994, 1997; Zakzanis, 2000) were not found in the present cohort of PPMS participants. As the presence of language deficits has been established in some patients with CPMS, it may be the case that, as with other areas of cognition, the language abilities of individuals with SPMS are more severely impaired than the language abilities of individuals with PPMS.

Previous research demonstrates that semantic fluency has a heightened sensitivity to MS-control comparisons than phonemic fluency (Beatty, 2002; Zakzanis, 2000). Accordingly, it is tangible that deficits in semantic fluency may have been missed in the present study or may have been unidentifiable in this small cohort. Indeed, Huijbregts et al. (2004) showed that participants with PPMS had reduced performance compared to controls on a semantic fluency task. Other examinations have found that MS participants have difficulty on tests of vocabulary and semantic knowledge (Laatu et al., 1999; Lethlean & Murdoch, 1997), and on language tests that rely on independent response initiation and speed (Wishart & Sharpe, 1997). Tests of these abilities were not specifically included in the present study.

Clinical implications

The findings of this preliminary case and group investigation suggest a potential language dysfunction in persons with PPMS. From a clinical perspective, validation of the present findings on a larger cohort of persons with PPMS is highly warranted to assist both in elucidating the extent of language deficits induced by PPMS and providing a direction for appropriate rehabilitation strategies. As evidenced in the present study, the TLC-E may represent a potentially useful assessment to define and characterize high-level language impairments in some patients with PPMS.

Conclusions

The results of the current preliminary study suggest that, although patients with PPMS may have preserved general language abilities, some individuals have the potential to present with mild impairments in meta-linguistic abilities. As only a limited number of participants with PPMS participated in the present investigation, the language abilities of a larger group of participants should be described in future research to determine the validity of the obtained results, both in terms of the sensitivity of language assessments and language outcomes. Additional assessments of speed of language processing, vocabulary, and semantic fluency may provide valuable information about language functions affected by PPMS. Comparison of language disorders presenting in PPMS, RRMS, and SPMS may also serve to further enhance understanding of the relationship between disease course and language impairment in MS.

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References

- Beatty, W. W. (2002). Fluency in multiple sclerosis: Which measure is best? *Multiple Sclerosis Journal*, 8, 261–264.
- Beatty, W.W., Goodkin, D. E., Beatty, P. A., & Monson, N. (1989). Frontal lobe dysfunction and memory impairment in patients with chronic progressive multiple sclerosis. *Brain and Cognition*, 11, 73–86.

- Beatty, W. W., Hames, K. A., Blanco, C. R., Paul, R. H., & Wilbanks, S. L. (1995). Verbal abstraction deficit in multiple sclerosis. *Neuropsychology*, 9, 198–205.
- Bennett, T., Ditmar, C., & Raubach, S. (1991). Multiple sclerosis: Cognitive deficits and rehabilitation strategies. *Journal of Cognitive Rehabilitation*, 5, 18–23.
- Brassington, J. C., & Marsh N. V. (1998). Neuropsychological aspects of multiple sclerosis. *Neuropsychology Review*, 8, 43–77.
- Comi, G., Filippi, M., Martinelli, V., Campi, A., Rodegher, M., Alberoni, M., Sirabian, G., & Canal, N. (1995). Brain MRI correlates of cognitive impairment in primary and secondary progressive multiple sclerosis. *Journal of Neurological Sciences*, 132, 222–227.
- Cook, M., Murdoch, B., Cahill, L., & Whelan, B. M. (2004). Higher-level language deficits resulting from left primary cerebellar lesions. *Aphasiology*, 18, 771–784.
- DeSonneville, L. M. J., Boringa, J. B., Reuling, I. E. W., Lazeron, R. H. C., Adèr, H. J., & Polman, C. H. (2002). Information processing characteristics in subtypes of multiple sclerosis. *Neuropsychologia*, 40, 1751–1765.
- FitzGerald, F. J., Murdoch, B. E., & Chenery, H. J. (1987). Multiple sclerosis: Associated speech and language disorders. *Australian Journal of Communication Disorders*, 15, 15–33.
- Foong, J., Rozewicz, L., Chong, W. K., Thompson, A. J., Miller, D. H., & Ron, M. A. (2000). A comparison of neuropsychological deficits in primary and secondary progressive multiple sclerosis. *Journal of Neurology*, 247, 97–101.
- Friend, K. B., Rabin, B. M., Groninger, L., Deluty, R. H., Bever, C., & Grattan, L. (1999). Language functions in patients with multiple sclerosis. *Clinical Neuropsychology*, 13, 7–94.
- Gaudino, E. A., Chiaravalloti, N. D., DeLuca, J., & Diamond, B. J. (2001). A comparison of memory performance in relapsingremitting, primary progressive and secondary progressive, multiple sclerosis. *Neuropsychiatry, Neuropsychology, and Behavioral Neurology*, 14, 32–44.
- Grossman, M., Robinson, K. M., Onishi, K., Thompson, H., Cohen, J., & D'Esposito, M. (1995). Sentence comprehension in multiple sclerosis. *Acta Neurologica Scandinavica*, 92, 324–331.
- Heaton, R. K., Taylor, M. J., & Manly, J. (2003). Demographic effects and use of demographically corrected norms with the WAIS-III and WMS-III. In D. S. Tulsky, D. H. Saklofske, G. J. Chelune, R. K. Heaton, R. J. Ivnik, R. Bornstein, et al (Eds.), *Clinical Interpretation of the WAIS-III and WMS-III.* (pp. 181–210). San Diego, CA: Academic Press.
- Huijbregts, S. C., Kalkers, N. F., de Sonneville, L. M., de Groot, V., Reuling, I. E., & Polman, C. H. (2004). Differences in cognitive impairment of relapsing remitting, secondary, and primary progressive MS. *Neurology*, 63, 335–339.
- Kaplan, E., Goodglass, H., & Weintraub, S. (1983). The Boston Naming Test. Philadelphia, PA: Lea & Febiger.
- Kraus, J.A., Schutze, C., Brokate, B., Kroger, B., Schwendemann, G., & Hildebrandt, H. (2005). Discriminant analysis of the cognitive performance profile of MS patients differentiates their clinical course. *Journal of Neurology*, 252, 808–813.
- Laatu, S., Hamalainen, P., Revonsuo, A., Portin, R., & Ruutiainen, J. (1999). Sematic memory deficit in multiple sclerosis: Impaired understanding of conceptual meanings. *Journal of Neurological Sciences*, 162, 152–161.
- Lethlean, J. B., & Murdoch, B. E. (1993). Language problems in multiple sclerosis. *Journal of Medical Speech-Language Pathology*, 1, 47–59.
- Lethlean, J. B., & Murdoch, B. E. (1994). Naming errors in multiple sclerosis: Support for a combined semantic/perceptual deficit. *Journal of Neurolinguistics*, 8, 207–223.
- Lethlean, J. B., & Murdoch, B. E. (1997). Performance of subjects with multiple sclerosis on tests of high-level language. *Aphasiology*, 11, 39–57.
- Lublin, F. D., & Reingold, S. C. (1996). Defining the clinical course of multiple sclerosis: Results of an international survey. National Multiple Sclerosis Society (USA) Advisory Committee

on Clinical Trials of New Agents in Multiple Sclerosis. *Neurology*, 46, 907–911.

- Mattis, S. (1988). *Dementia Rating Scale*. Odessa, FL: Psychological Assessment Resources.
- Palace, J. (2003). The diagnosis of primary progressive multiple sclerosis. *Journal of Neurological Sciences*, 206, 145–152.
- Spreen, O., & Benton, A. L. (1969). Neurosensory Center Comprehensive Examination for Aphasia. Victoria, Canada: University of Victoria.
- Thompson, A. J., Polman, C. H., Miller, D. H., McDonald, W. I., Brochet, B., Filippi, M. et al. (1997). Primary progressive multiple sclerosis. *Brain*, 120, 1085–1096.
- Thompson, A. J., Montalban, X., Barkhof, F., Brochet, B., Filippi, M., Miller, D. H., et al. (2000). Diagnostic criteria for primary progressive multiple sclerosis: A position paper. *Annals of Neurology*, 47, 831–835.
- Vukusic, S., & Confavreux, C. (2003). Primary and secondary progressive multiple sclerosis. *Journal of Neurological Sciences*, 206, 153–155.
- Wachowius, U., Talley, M., Silver, N., Heinze, H. J., & Sailer, M. (2005). Cognitive impairment in primary and secondary

progressive multiple sclerosis. Journal of Clinical and Experimental Neuropsychology, 27, 65-77.

- Weinshenker, B. G., Bass, B., Rice, G. P., Noseworthy, J., Carriere, W., Baskerville, J., et al. (1989) The natural history of multiple sclerosis: A geographically based study. I. Clinical course and disability. *Brain*, 112, 133–146.
- Whitaker, J. N., & Mitchell, G. W. (1997). Clinical features of multiple sclerosis. In C. S. Raine, H. F. McFarland, & W. W. Tourtellote (Eds.), *Multiple sclerosis: Clinical and pathogenic basis.* (pp. 3–19). London: Chapman and Hall.
- Wiig, E. H., & Secord, W. (1989). Test of Language Competence Expanded. New York, NY: Psychological Corporation.
- Wishart, H., & Sharpe, D. (1997). Neuropsychological aspects of multiple sclerosis: a quantitative review. *Journal of Clinical and Experimental Psychology*, 19, 810–824.
- Wolinsky, J. S. (2003). The diagnosis of primary progressive multiple sclerosis. *Journal of Neurological Sciences*, 206, 145–152.
- Zakzanis, K. K. (2000). Distinct neurocognitive profiles in multiple sclerosis subtypes. Archives of Clinical Neuropsychology, 15, 115–136.