The relationship between non-orthographic language abilities and reading performance in chronic aphasia: An exploration of the primary systems hypothesis

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Abstract

Purpose: This study investigated the relationship between non-orthographic language abilities and reading in order to examine assumptions of the primary systems hypothesis and further our understanding of language processing post-stroke.

Method: Performance on non-orthographic semantic, phonologic, and syntactic tasks, as well as oral reading and reading comprehension tasks was assessed in 43 individuals with aphasia. Correlation and regression analyses were conducted to determine the relationship between these measures. Additionally, ANOVA analyses examined differences within and between reading groups (within normal limits, phonological, deep, or global alexia).

Results: Results showed non-orthographic language abilities were significantly related to reading abilities. Semantics was most predictive of regular and irregular word reading, while phonology was most predictive of pseudohomophone and nonword reading. Written word and paragraph comprehension were primarily supported by semantics, whereas, written sentence comprehension was related to semantic, phonologic, and syntactic performance. Finally, severity of alexia was found to reflect severity of semantic and phonologic impairment.

Conclusions: Findings support the primary systems view of language by showing non-orthographic language abilities and reading abilities are closely linked. This preliminary work requires replication and extension; however, current results highlight the importance of routine, integrated assessment and treatment of spoken and written language in aphasia.

Introduction

Aphasia, an acquired difficulty with language processing, results from damage to the language-dominant hemisphere of the brain, most commonly following left-hemisphere stroke. Although aphasia is not a uniform disorder, people with aphasia (PWA) tend to present with pervasive, multi-modal language impairment characterized by deficits in auditory comprehension, spoken production, reading, and writing, to varying degrees. Much of aphasia research and clinical management has been directed towards understanding and rehabilitating spoken language impairment with less emphasis on written language impairment (Knollman-Porter, Wallace, Hux, Brown, & Long, 2015; Thiel, Sage, & Conroy, 2015). However, with today's increased reliance on written communication (e.g., emails, text messages, social media, etc.), PWA are expressing a greater interest in improving their reading and writing, and more research devoted to written language is warranted (Thiel et al., 2015; Webster et al., 2013). Surprisingly, the relationship between co-occurring spoken and written language abilities in aphasia has received little attention, despite the fact that better understanding of this relationship may enhance our understanding of language processing and inform aphasia rehabilitation.

This project is focused on the reading (versus spelling) aspect of written language and, more specifically, how acquired reading impairment (alexia) relates to spoken language impairment in PWA. The nature of acquired alexia has been extensively studied for over a century, and the interested reader is referred to work by esteemed alexia researchers, such as Cherney (2004), Beeson, Rising, Kim, and Rapscak (2010), Rapp, Folk, and Tainturier (2001), Friedman (1996), Leff and Starrfelt (2014), Coltheart, Rastle, Perry, Langdon, and Ziegler (2001), Plaut (1996), and Lambon Ralph and Patterson (2007), to name only a few. It is important to note that alexia research has shed great light on the process of reading by revealing that reading can be impaired in many ways, including peripherally (i.e., at the level of visual processing) and centrally (i.e., via damage to linguistic processes such as semantics or phonology). Moreover, this work has shown that numerous linguistic variables, such as word length, frequency, imageability, part of speech, lexicality, and spelling-sound regularity affect reading, and differential patterns of performance related to manipulation of these linguistic parameters help to characterize the nature and underlying basis for reading impairment in a given individual.

The co-occurrence of alexia and aphasia is now widely recognized. Although there can be a dissociation between written and spoken language abilities (Howard & Nickels, 2005), the majority of individuals with left hemisphere stroke-induced aphasia present with alexia (Brookshire, Wilson, Nadeau, Gonzalez-Rothi, & Kendall, 2014; Leff & Starrfelt, 2014; Luzzatti et al., 2006; Webb & Love, 1983). Unfortunately, in aphasia rehabilitation a person's alexia is typically examined (and treated) separately from his/her spoken language impairment (Leff & Starrfelt, 2014).

This disconnect between aphasia and alexia research, might be related to the traditional notion that written and spoken language are supported by functionally unrelated neural processes (Lambon Ralph & Patterson, 2007), as commonly depicted in popular language models. Spoken language models (e.g., Dell, Schwartz, Martin, Saffran, & Gagnon, 1997) typically do not consider the relevance of orthographic abilities, and likewise, written language models (e.g., Coltheart et al., 2001) do not address the impact spoken language abilities may have on reading and spelling abilities or the potential for core skills that allow language to be shared across modalities. Some models (e.g., Rapcsak & Beeson, 2000) illustrate both spoken and written language processing; however, spoken and written language are still depicted as relying on independent components specific to each language modality.

Contrary to the view of distinct spoken and written language systems, a parallel-distributed processing (PDP) connectionist model of single-word processing (Plaut, 1996; Seidenberg & McClelland, 1989) promotes the view that spoken and written word processing involves synchronized activation of semantic, phonologic, and orthographic units, with word knowledge existing as a learned pattern of neural activity that resides in the connections between these distributed language units. Since word processing results from semantic, phonologic, and orthographic information simultaneously interacting and settling into a learned pattern of activation, there are no proposed lexicons or grapheme-phoneme rule systems, as is commonly assumed in classic, sequential views of language processing. The connectionist/triangle model comprises three pathways: semantics-phonology, phonology-orthography, and orthography-semantics (see Figure 1) that contribute to language processing, with a division of labor developing as the language system becomes more efficient. That is, certain language units have more input to the final activation pattern, depending on the specific language task or stimulus. For example, when reading irregularly spelled words (e.g., chef), more semantic contribution is expected, whereas when reading unfamiliar or nonwords (e.g., splooch), greater phonological contribution is critical.

INSERT FIGURE 1 ABOUT HERE

Based on this connectionist account of language processing, the primary systems hypothesis (PSH) emerged and proclaims that three connected primary neural systems (visual, semantic, phonologic; see Figure 1) interact to collectively contribute to the reading of all words, and importantly, that disruption to one or more of these modality-independent, primary systems is responsible for reading impairment (Lambon Ralph & Patterson, 2007; Patterson & Lambon Ralph, 1999; Woollams, 2014). This notion stems from the PSH's evolutionary and developmental standpoint that reading is (a later acquired) part of the larger language system, as opposed to a skill independent from other language abilities (Woollams, 2014). The PSH assumes that later-acquired written language develops from and relies on the same cognitive mechanisms that support earlier-acquired spoken language abilities. Therefore, significant relationships between spoken and written language impairments are expected (Woollams, 2014).

In the context of the PSH, Crisp and Lambon Ralph (2006) depicted the expected relationships between impaired oral reading of content words and semantic and phonologic impairment. As illustrated in Figure 2, an individual's reading ability (or inability) reflects the status of his or her primary language systems. *Normal reading* ability is associated with intact semantic and phonological processing abilities. *Surface alexia*, which is characterized by poor reading of irregularly spelled words with (mostly) intact reading of regularly spelled words (regularity reading effect), is proposed to stem from underlying semantic impairment. This account differs from the traditional dual-route explanation (Coltheart, Curtis, Atkins, & Haller, 1993; Coltheart et al., 2001) of surface alexia resulting from an exclusive impairment to a direct, lexical reading route. *Phonological alexia*, which is characterized by poor nonword reading compared to relatively intact real word reading (lexicality reading effect), is purported to result from a high degree of phonological impairment. This explanation from Crisp and Lambon Ralph differs from Coltheart and colleagues' dual-route perspective of a damaged indirect, sublexical reading route and selectively impaired grapheme-to-phoneme rule system. *Deep alexia*, which is commonly accepted as a severe form of phonological alexia (Crisp & Lambon Ralph, 2006; Crisp, Howard, & Lambon Ralph, 2011; Friedman, 1996; Glosser & Friedman, 1990), is hypothesized to arise from some semantic impairment, in addition to severe phonological impairment. This perspective held by Crisp and Lambon Ralph (2006) varies from the classic view of deep alexia resulting from damage to both lexical and sublexical reading-specific routes. Finally, *global alexia* is proposed to be associated with severe underlying semantic and phonological impairment, which is thought to account for the abolished reading ability that defines the syndrome. Figure 2 pertains only to central alexias and therefore pure alexia (letter-by-letter reading) is not pictured; however, the PSH proposes this type of peripheral alexia stems from an impaired ability to process visually complex stimuli, as opposed to a selective disorder of slow and labored visual analysis of letters (Lambon Ralph & Patterson, 2007).

INSERT FIGURE 2 ABOUT HERE

Empirical support for the PSH and PDP theory, jointly referred to as the "triangle model/primary systems hypothesis" by Crisp and Lambon Ralph (2006, p. 358), comes not only from acquired alexia, but also behavioral work in developmental dyslexia, as well as neuroimaging findings.

Neuroimaging data show the process of reading engages distributed neural regions. Brunswick (2010) explains that the functional neuroanatomy of reading includes brain regions associated with visual processing, semantic processing, and phonological processing. Orthographic (letter) processing is thought to primarily rely on modified and "recycled" neurons in the left posterior occipitotemporal sulcus (i.e., visual word form area) that originally evolved for other visual processing purposes (Dehaene & Cohen, 2010). Visual letter processing then works in concert with anterior (e.g., left inferior frontal cortex) and posterior (e.g., left supramarginal gyrus and anterior inferior parietal cortex) language systems to result in skilled reading (Brunswick, 2010; Leff and Starrfelt, 2014). These findings align with the PSH notion that reading is “parasitic” (Woollams, 2014, p. 8) on preexisting cortical systems and relies on an interaction between visual and language systems that are also involved in other functions.

The developmental dyslexia literature also supports the idea that reading emerges from and is dependent upon other language skills. Spoken language abilities (phonology, semantics, syntax) are known to directly influence children’s reading development (Nation, 2007; Perfetti, Landi, & Oakhill, 2007; Wagner & Torgesen, 1987) and impaired oral language is thought to be at the core of reading impairment (Hulme & Snowling, 2014). Furthermore, children with specific language impairment present with reading difficulties, in addition to the more recognized oral language difficulty, and likewise children with developmental dyslexia are known to have phonological impairment, in addition to reading impairment (Bishop & Snowling, 2004), supporting the inherent connection between spoken and written language abilities proposed by the PSH.

Although the concept of interrelated spoken and written language impairment is generally accepted in the developmental dyslexia field (Hulme & Snowling, 2014), support for this relationship in adults with acquired reading impairment is more limited. Emerging evidence comes from studies investigating the relationship between orthographic impairment and non-orthographic visual, semantic, or phonologic impairment. For example, research has shown that individuals with pure alexia demonstrate impaired identification of complex visual stimuli other than letters, suggesting damage to central visual processes (Lambon Ralph & Patterson, 2007). Similarly, individuals with surface alexia have displayed trouble processing irregular stimuli beyond irregularly spelled words, including irregular past tense verbs (Patterson, Lambon Ralph, Hodges, & McClelland, 2001). Furthermore, semantic performance has been significantly correlated with irregular word reading in this population (Woollams et al., 2007), indicating general semantic impairment. Finally, multiple studies have shown that individuals with phonological-deep alexia demonstrate impaired phonological processing (Crisp & Lambon Ralph, 2006; Jefferies et al., 2007; Rapcsak et al., 2009). Moreover, these studies found that phonological abilities significantly predicted oral reading and/or spelling abilities, supporting an underlying phonological impairment.

In addition to phonological impairment in phonological-deep alexia/agraphia, Rapcsak et al. (2009, p. 581) stated "it remains to be determined, however, whether the proposed continuum is best characterized by the severity of the phonological deficit, the degree of semantic impairment, or a combination of both factors". Two studies that simultaneously examined phonologic and semantic performance in alexia found each of these language domains underpinned reading (Crisp & Lambon Ralph, 2006) or both reading and spelling (Henry et al., 2012). In particular, in individuals with primary progressive aphasia, Henry et al. (2012) reported performance on non-orthographic semantic measures (i.e., picture category association, synonym judgment, spoken word to picture matching, picture naming) related most strongly with irregular word reading and spelling, while performance on non-orthographic phonologic measures (i.e., rhyme judgment, minimal pair discrimination, phoneme manipulation) related most strongly with nonword reading and spelling. Together, these findings support the PSH claim that acquired written and spoken language impairments reflect damage to shared semantic and phonologic processes.

The findings summarized above from the neuroimaging, dyslexia, and alexia literatures advocate for the PSH; however, the PSH is relatively new (in the long history of reading research) and not widely endorsed. Therefore, additional work is warranted to further examine the relationships proposed by the PSH and to better understand acquired reading impairment in aphasia. The PSH has not yet been tested in a large, heterogeneous sample of individuals with chronic, stroke-induced aphasia. In previous PSH studies, sample sizes were small and inclusion was limited to individuals with aphasia with phonological or deep alexia (Crisp & Lambon Ralph, 2006), left peri-sylvian damage (Rapcsak et al., 2009), or primary progressive aphasia (Henry et al., 2012). Furthermore, reading (or spelling) was measured solely at the single word level. Thus, we remain uncertain about the relationship between non-orthographic language abilities and written language skills beyond the single word level in a large group of PWA.

The current study aimed to examine the PSH to better understand the relationship between orthographic language performance, specifically oral reading and silent reading comprehension, and non-orthographic language performance in a diverse group of PWA. To that end, the following research questions were posed:

1. What is the relationship between non-orthographic semantic and phonologic abilities and oral reading of regular words, irregular words, pseudohomophones, and nonwords?
2. What is the relationship between non-orthographic semantic, phonologic, and syntactic abilities and reading comprehension at the single-word, sentence, and paragraph level?
3. What is the relationship between degree of reading impairment (alexia severity) and non-orthographic language performance?

In accordance with previous findings (Crisp & Lambon Ralph, 2006; Henry et al., 2012; Jefferies et al., 2007; Rapcsak et al., 2009) and assumptions of the PSH and PDP theory, non-orthographic semantic and phonologic measures were hypothesized to significantly predict oral reading performance in our sample, with phonology being most related to nonword reading and semantics most related to irregular word reading. Regarding reading comprehension, it was expected that semantics and phonology would predict written word comprehension to the greatest extent with syntax being most predictive of written sentence comprehension. Finally, degree of reading impairment was anticipated to be directly linked to degree of semantic and phonologic impairment with the most impaired readers (e.g., global alexia) demonstrating the most impaired non-orthographic language abilities and the least impaired readers (e.g., within normal reading limits) showing the most intact non-orthographic language performance.

Method

Participants

Forty-seven PWA were recruited through the University of Washington (UW) and Northwest Aphasia Repository Databases and participated in the study. To be included, participants presented with aphasia and were at least six months post onset of a left-hemisphere stroke. Additionally, participants obtained at least a high school education and spoke English as their primary language. Presence of aphasia was determined by performing below the cut-off scores on at least two of the following subtests from the Comprehensive Aphasia Test (CAT; Swinburn, Porter, & Howard, 2004): Comprehension of Spoken Language, Naming, Repetition, and Spoken Picture Description (See Supplementary Appendix A online for individual and group performance on the CAT).

Individuals with a history of developmental dyslexia or neurological diagnosis other than left-hemisphere stroke were excluded. PWA were also excluded for failing vision or hearing screenings. Moreover, five speech-language pathologists (SLPs) in the UW aphasia research lab reviewed two video-recorded spoken production tasks for each participant and, based on consensus, identified and excluded those with severe motor speech impairment. Spoken production was evaluated for behaviors associated with apraxia of speech (AOS) (i.e., slow rate, prolonged segment durations, intrusive schwa, distorted substitutions, abnormal stress) and dysarthria (i.e., hyper/hypo-nasality, impaired vocal quality, muscle weakness, imprecise articulation, reduced breath support) (Duffy, 2005).

Forty-three of the 47 enrolled participants met the study inclusion/exclusion criteria and were included in the final analyses. Two participants were excluded due to lesion location other than left cerebral hemisphere and two were excluded due to severe motor speech impairment. The final sample consisted of 25 males and 18 females whose average age was 62.12 years (*SD* = 11.39; range = 31-92), average time post-stroke onset was 69.16 months (*SD* = 45.42; range = 8-216), and average education was 15.72 years (*SD* = 2.62; range =12-22). See Supplementary Appendix A online for individual demographic information.

Procedure

Testing Administration. Each participant was tested over 2-3 days in a quiet room. For tasks that involved an auditory stimulus, the volume was adjusted for each participant to ensure the stimulus was comfortably audible. For oral reading tasks, participants wore a head mounted microphone, and responses were audio-video recorded for later scoring, error, and reliability analyses. For all other language tasks, responses were non-verbal and recorded on paper by the examiner.

Experimental Testing. Semantic, phonologic, syntactic, oral reading, and silent reading comprehension abilities were assessed via numerous tests (See Table 1). These tests were chosen due to their similarity to tests administered in other studies that have investigated the relationship between reading and primary language systems (Crisp & Lambon Ralph, 2006; Henry et al., 2012; Jefferies et al., 2007; Rapcsak et al., 2009). However, the inclusion of syntactic and silent reading comprehension measures is novel to this study and authors selected these measures from commonly used aphasia standardized test batteries. The semantic, phonologic, and syntactic tests in this study did not include any letters (i.e., no orthography), and furthermore, all involved non-verbal responses to reduce the impact of any motor speech impairment. Multiple measures for each language domain were administered in an effort to create variation in difficulty to help prevent floor or ceiling effects and to better capture the language domain since no single task can encompass the entire domain. For each participant, the order of presentation (i.e., semantics, phonology, syntax, oral reading, and silent reading comprehension) and the order of tasks within each language domain were presented in random order.

INSERT TABLE 1 ABOUT HERE

As a way to justify the a priori grouping of tests and the creation of composite scores (see below), a principal components analysis (PCA) was performed for each language domain to determine whether or not the tests in that domain loaded on only one component and represented a single factor. For each language domain, the correlation matrix was inspected to ensure that the tests within each domain had at least one correlation coefficient greater than 0.3, that the overall Kaiser-Meyer-Olkin (KMO) measure was greater than 0.6, and that Bartlett's test of sphericity was statistically significant, indicating that the data was appropriate for a PCA (Laerd Statistics, 2015). Only one component, or factor, with an eigenvalue greater than one was extracted in each of the five PCAs. In particular, the semantic tests loaded on one factor with an eigenvalue of 2.80 that explained 69.99% of the variance, and the phonologic tests loaded on one factor with an eigenvalue of 2.26 that explained 56.50% of the variance. An eigenvalue of 2.54 explained 84.68% of the variance in the syntactic tests, and an eigenvalue of 3.41 explained 85.35% of the variance in the oral reading tests. Finally, the silent reading comprehension tests loaded on one factor with an eigenvalue of 6.45 that explained 71.70% of the variance. Supplementary

Appendix B online contains correlational values between each of the tests in each language domain, for the interested reader.

Scoring. The items on each test received a raw accuracy score (correct = 1; incorrect = 0). For the oral reading tasks, responses with speech sound distortions were scored as correct. Phonological errors (e.g., sound omissions, substitutions, transpositions, and additions), semantic errors, and non-responses were scored as incorrect. It is important to acknowledge that for participants with aphasia and mild-moderate AOS, distinguishing phonological from phonetic errors is inherently difficult. For this reason, individuals with severe AOS were excluded and sound distortions permitted, as previously mentioned, in an effort to minimize scoring errors.

For all tasks, self-corrections were permitted and the participant's final response was scored. The total raw score for each test was converted into a z-score (z-score = (individual score – group mean)/*SD*) to allow for a standard comparison across the various measures that differed in language domain, number of items, and/or level of difficulty. The z-score values were used to create composite scores for each participant in each language domain with composite scores consisting of averaged z-scores from each language domain. For example, each participant's semantic composite score was derived by averaging his/her four z-scores from the four semantic tasks.

Calculating Alexia Subtypes. A reading profile (i.e., surface, phonological, deep, or global alexia, or within normal reading limits) was calculated for each participant so the relationship between reading ability and non-orthographic language abilities could be further explored. Alexia type was determined for each participant based on his/her oral reading accuracy and the types of oral reading errors produced. Similar to Brookshire, Conway, Hunting Pompon, Oelke, and Kendall (2014), oral reading accuracy was determined by calculating each participant's 95% reading accuracy confidence interval (CI) for reading of regular words, irregular words, all real words, pseudohomophones, and nonwords. These calculations were made using the following formula: *95% CI = participant's average score +/- (1.96 x SEM)*. For example, Participant 39 read real words with 80% accuracy so her real word 95% CI = .80 +/ - (1.96 x .03) = .80 +/-.06 for a real word accuracy score range of 86%-74%. The 95% CI was calculated in an effort to provide a conservative estimate of reading ability compared to using the average percent correct score.

Trained research assistants viewed audio-video recordings of each participant's oral reading in order to determine the types of oral reading errors produced. Oral reading errors were classified into one of the following seven error types: regularization reading error *(*e.g., reading "pint" to rhyme with "mint"), visual/phonological reading error (e.g., "single" for "signal"), lexicalization of nonword reading error (e.g., "fig" for "flig"), semantic reading error (e.g., "boat" for "yacht"), unrelated real word reading error (e.g., "bed" for "count"), unrelated nonword reading error (e.g., "eebee" for "gang"), or omission reading error (e.g., "I don't know").

After calculating oral reading accuracy (95% CI) for each word type (i.e. regular, irregular, pseudohomophone, and nonwords) and oral reading errors, participants' reading profiles were determined. *Surface alexia* was defined by presence of regularization reading errors, as well as presence of a regularity reading effect (e.g., better reading of regularly spelled words than irregularly spelled words), which was defined by non-overlapping regular and irregular word reading ranges (95% CIs) and non-overlapping nonword and irregular word reading ranges, with regular and nonword reading being superior to irregular word reading. *Phonological alexia* was characterized by predominant lexicalization or visual/phonologic errors, as well as presence of a lexicality reading effect (i.e., better reading of real words compared to nonwords) which was defined by non-overlapping real word and nonword reading accuracy ranges with real word reading being superior to nonword reading. Moreover, to account for real word reading ability in phonological alexia being quite variable, yet typically still relatively well preserved (Farah et al., 1996; Patterson & Lambon Ralph, 1999), the high end of the real word 95% CI was defined as being at least 60% accurate. Given that overall greater reading impairment and the production of semantic reading errors are the hallmark features of individuals on the deep end of the phonological-deep continuum (Crisp & Lambon Ralph, 2006; Lambon Ralph & Patterson, 2007), *deep alexia* was characterized by overall reading being 50% accurate or less with the presence of semantic errors. Additionally, the high end of the real word 95% CI was less than 60% accurate and the high end of the nonword 95% CI was less than 30% accurate to ensure a lexicality effect was present. *Global alexia*, abolished or near abolished reading ability, was defined as accurately reading 10% or less of the oral reading stimuli (reading less than 11/112 words). Participants who performed within two standard deviations of normal controls on the regular words (*M* = 99.56%; *SD* =2.17%), irregular words (*M* =98.06%; *SD* = 4.57%), nonwords (*M* = 95.74%; *SD* = 7.30%), and pseudohomophones (*M* = 79.30; *SD* = 18.24%) were considered to read *within normal limits (WNL).* Finally, participants who did not read WNL and did not meet the defined alexia subtype criteria described above, were labeled as having an "unclassified" reading impairment profile.

Reliability. Intra- and inter-rater reliability for reading accuracy was performed for 25% of each participant's oral reading data. A randomly selected 25% of each word type (i.e., 10 regular, 10 irregular, 5 nonwords, and 3 pseudohomophones) was re-analyzed. The overall proportion of agreement between the initial and intra-rater accuracy ratings was .97 and the proportion of agreement between two raters was .96. In order to account for chance agreement, Cohen's Kappa (*k*) was also calculated, with values of .93 ($p < .001$) and .91 ($p < .001$) for the intra- and inter-rater accuracy agreements, respectively. Intra- and inter-rater reliability for classifying reading error types was performed on the same 25% of stimuli for each participant. The overall proportion of agreement between the initial and intra-rater error code ratings was .95 and the proportion of agreement between the two raters was .92. Cohen's Kappa values of .90 ($p < .001$) and .86 ($p < .001$) were found for the intra- and inter-rater error coding agreements, respectively.

Statistical Analysis. To address research question 1 (RQ1) and determine the relationship between oral reading abilities and non-orthographic semantic and phonologic abilities, oral reading z-scores were correlated with phonological and semantic z-scores. Additionally, five standard multiple linear regression (MLR) models with simultaneous entry were performed using the phonology and semantic composite scores as predictors of 1) overall oral reading, 2) regular word reading, 3) irregular word reading, 4) pseudohomophone reading, and 5) nonword reading to determine to what extent phonology and semantics account for the variance in oral reading accuracy of different word types.

To address RQ2 and determine the relationship between silent reading comprehension abilities and non-orthographic language abilities, reading comprehension z-scores were correlated with semantic, phonologic, and syntactic z-scores. Then, four standard MLR analyses with simultaneous entry were completed using semantic, phonologic, and syntactic composite scores as predictors of 1) overall reading comprehension, 2) single-word reading comprehension, 3) sentence-level reading comprehension, and 4) paragraph-level reading comprehension to determine to what extent these predictors account for the variance in silent reading comprehension, and at what linguistic level.

To address RQ3 and determine the relationship between non-orthographic semantic and phonologic performance and alexia type, correlations were calculated between performance on semantic and phonologic measures and selected hallmark alexia symptoms. Specifically, hallmark alexia features that were possible to calculate from the oral reading stimuli used in this study included a regularity effect (i.e., difference between percent correct regular and irregular word reading), a lexicality effect (i.e., difference between percent correct real word and nonword reading), and a frequency effect (i.e., difference between percent correct reading high versus low frequency words), as well as proportion of reading errors (i.e., regularization, visual/phonologic, lexicalization, semantic, omission, and unrelated reading errors). Furthermore, semantic and phonological performance within and between the alexia groups was examined.

Statistical comparisons in this study were motivated by a priori hypotheses and therefore an alpha value of 0.05 (two-tailed test) was used to indicate statistical significance for all analyses. The reader might interpret findings cautiously in light of multiple comparisons. The authors' decision not to correct for multiple comparisons is shared by other researchers in the field with pre-planned comparisons and small sample sizes (Ash et al., 2011; Minkina, Martin, Spencer, & Kendall, 2018). This decision attempts to balance the probability of making type I and type II errors given the probability of making a type I error is smaller for a priori than post hoc multiple comparisons (Ash et al., 2011) and the likelihood of making a type II error is inflated when correcting for multiple comparisons (Perneger, 1998).

Results

<u>RQ 1: Non-orthographic semantic and phonologic performance and oral reading performance</u>

Descriptive Statistics. As expected in a large, diverse group of PWA there was a wide range in language ability. Group performance on the semantic, phonologic, and oral reading tests is presented in Table 2.

INSERT TABLE 2 ABOUT HERE

Correlation Results. The semantic and phonologic composite scores were both significantly associated with the oral reading composite score (r = .70, p < .01; r = .65, p <.01, respectively). The strongest semantic composite relationship was with reading of irregular words (r = .71, p < .01), then regular words (r = .67, p < .01), pseudohomophones (r = .63, p < .01), and the lowest correlation was with nonwords (r = .56, p < .01). This same relationship pattern (irregular > regular > pseudohomophone > nonword) was seen for each of the four semantic tasks comprising the semantic composite. The strongest phonologic composite relationship was with pseudohomophone reading (r = .71, p < .01), followed by nonword reading (r = .57, p <.01), then regular and irregular word reading (r =. 55, p < .01). This pattern (pseudohomophones > nonwords > real words) was seen for all of the phonological tasks that comprised the phonology composite, except minimal pair discrimination showed the strongest relationship with real word reading followed by pseudohomophones and nonwords.

Regression Results. Table 3 illustrates the results of five MLR models with the semantic and phonologic composite scores entered simultaneously as the predictors and 1) total oral reading accuracy (all word types), 2) regular word accuracy, 3) irregular word accuracy, 4) pseudohomophone accuracy, and 5) nonword accuracy entered as the respective outcome variables. For each model, the assumption of linearity was confirmed via inspection of partial regression plots. The tolerance collinearity statistic was 0.64 for both predictors, indicating that the relationship between the semantic and phonologic composites is not likely to interfere with the analysis (multicollinearity) given a tolerance value greater than 0.1 is generally acceptable (Laered Statistics, 2015). The predictors together accounted for a statistically significant amount of the variance for total oral reading (R^2 = .57, p <.001), regular words (R^2 = .49, p <.001), irregular words (R^2 = .53, p <.001), pseudohomophones (R^2 = .57, p <.001), and nonwords (R^2 = .40, p <.001).

When phonology was held constant, semantics was found to have a unique effect on all word types. Specifically, if performance on the semantic composite improves one *SD*, there is an estimated mean increase of 0.54 *SD* on total reading (p < .001), 0.64 *SD* on regular words (p < .001), 0.71 *SD* on irregular words (p < .001), 0.38 *SD* on pseudohomophones (p < .05), and 0.40 *SD* on nonwords (p < .05). Holding performance on semantics constant, there is an estimated mean increase of 0.44 *SD* on total oral reading (p <.05), a 0.69 *SD* increase on pseudohomophone reading (p < .001), and a 0.50 *SD* increase on nonword reading (p < .05) for each *SD* gain on the phonology composite score.

INSERT TABLE 3 ABOUT HERE

RQ2: Non-orthographic language performance and silent reading comprehension performance

Descriptive Statistics. The participants' performance on the non-orthographic language tests and silent reading comprehension tests is summarized in Table 4.

INSERT TABLE 4 ABOUT HERE

Correlation Results. The semantic, syntactic, and phonologic composite scores were each significantly associated with the silent reading comprehension composite score (r = .82, p < .01; r = .75, p <.01, r = .65, p <.01, respectively). The strongest semantic composite relationship was with sentence-level written comprehension (r = .83, p < .01), then single words (r = .79, p < .01), and paragraphs (r = .72, p < .01). The auditory synonym judgment and comprehension of verbs and adjectives tasks showed stronger relationships with overall reading comprehension (r = .76, p < .01, r =.75, p <.01, respectively) compared to the picture association and word-to-picture matching tasks (r = .64, p < .01; r =.57, p <.01, respectively).

The syntactic composite was most strongly correlated with reading comprehension of sentences (r = .84, p < .01), and showed lower correlational values with comprehension of written words (r = .66, p < .01) and paragraphs (r = .64, p < .01). Of the syntactic tasks, comprehension of locative phrases had the highest correlation with reading comprehension (r = .74, p < .01). The strongest phonologic composite relationship was with written sentence comprehension (r = .69, p < .01), then paragraphs (r = .60, p < .01), and single words (r = .55, p < .01). The minimal pair discrimination task showed the strongest correlation with overall reading comprehension (r =. 63, p < .01), followed by nonword rhyme judgment (r =. 47, p < .01), phoneme manipulation (r = .42, p < .01), and real word rhyme judgment (r =. 40, p < .01).

Regression Results. Table 5 illustrates results of four MLR models with semantic, phonologic, and syntactic composite scores entered simultaneously as predictors and 1) overall reading comprehension, 2) written word comprehension, 3) written sentence comprehension, and 4) written paragraph comprehension entered as the outcome variables. For each model, the assumption of linearity was confirmed via inspection of partial regression plots. The tolerance collinearity statistic values for the predictors were all greater than 0.1 (semantics = 0.37, phonology = 0.57, and syntax = 0.34), indicating that the relationship between the predictors is not likely to interfere with the analysis (Laered Statistics, 2015). The predictors together accounted for a statistically significant amount of the variance for overall reading comprehension (R^2 = .72, p <.001), single word reading comprehension (R^2 = .63, p <.001), sentence-level reading comprehension (R^2 = .81, p <.001), and paragraph-level reading comprehension (R^2 = .57, p <.001).

Semantics had a unique effect on all reading comprehension levels. Specifically, there is an estimated mean increase of 0.71 *SD* on written word comprehension (p < .001), 0.39 *SD* increase on written sentence comprehension (p < .01), and 0.54 *SD* increase on written paragraph comprehension (p < .001) when performance increases one *SD* on the semantic composite. Phonology was shown to be uniquely predictive of sentence-level reading comprehension (p < .05) with an estimated mean increase of 0.23 *SD* on written sentence comprehension for each *SD* gain on the phonologic composite score. Syntax was also uniquely predictive of written sentence comprehension (p <.01) with an estimated increase of 0.41 *SD* for individuals improving one *SD* on the syntactic composite.

INSERT TABLE 5 HERE

RQ3: Relationship between semantic and phonologic impairment and alexia

Reading profiles. Thirty-seven of the 43 (86%) participants were identified to have alexia. Twenty-three participants (53%) met the phonological alexia classification, four participants (9%) were classified with deep alexia, five participants (12%) were classified with global alexia, no participants displayed surface alexia symptoms, and six participants (14%) performed within normal reading limits (WNL). Five participants (12%) demonstrated an alexia that could not be classified (CNC) according to the study criteria. One-way ANOVAs revealed no statistically significant differences in age (p = .20), education (p =.11), or time post-stroke onset (p =.64) between the reading groups.

Correlation Results. Correlations between non-orthographic semantic and phonologic abilities and hallmark alexia symptoms revealed that semantic performance had significant associations with regularity and frequency reading effects. As performance on the semantic composite improved, regularity and frequency effects both decreased (*r* = -.30, p <.05; *r* = -.31, p <.05, respectively). With regard to reading error types, as semantic abilities increased, the frequency of regularization (*r* = .32, p <.05), lexicalization (*r* = .37, p <.05), and visual-phonologic reading errors (*r* = .41, p <.01) also increased, while the frequency of omissions (*r* = -.39, p <.01), and unrelated real word reading errors (*r* = -.55, p <.01) decreased. The phonology composite was not significantly associated with regularity, lexicality or frequency effects; however, with regard to reading error types, as phonologic performance increased so did frequency of visual-phonologic errors (*r* = .39, p <.01), while frequency of omissions and unrelated real word errors decreased (*r* = -.39, p <.01; (*r* = -.31, p <.05, respectively).

Alexia group comparisons. Table 6 reports non-orthographic semantic and phonologic performance per reading group and shows the less impaired readers (i.e., WNL and phonological alexia) were the most accurate and performed above the group average (i.e., z-score above "0") on both semantic and phonologic tasks, whereas the more impaired readers (i.e., deep and global alexia) were the least accurate and performed below the group average (i.e., z-score below "0"). ANOVA analyses were conducted to determine if these differences were statistically significant.

INSERT TABLE 6 ABOUT HERE

Between-group comparisons. A one-way ANOVA revealed semantic performance was statistically significantly different between the reading groups (F (3, 34) = 6.78, p < .01, η^2 = 0.37). Planned follow-up t-test comparisons showed the WNL group demonstrated significantly better semantics compared to the deep alexia group (t (8) = 4.36, p =.002) and global alexia group (t (9) = 5.02, p =.001). The phonological alexia group displayed significantly better semantics than the global alexia group (t (26) = 3.53, p =.002) There were no significant differences between the phonological and deep alexia groups (p=.06), WNL and phonological alexia groups (*p* =.54) and the deep and global alexia groups (*p* = .17) on semantic performance (See Figure 3).

A subsequent one-way ANOVA showed phonologic performance was also statistically significantly different between the reading groups (F (3, 34) = 3.78, p =.02, η^2 = 0.23). Planned follow up t-test comparisons revealed the WNL group demonstrated better phonology than the deep alexia group (t (8) = 2.57, p =.03) and global alexia group (t (9) = 2.89, p =.02). The phonological alexia group demonstrated superior phonology over the global alexia group (t (26) = 2.25, p =.03). There were no significant phonological differences between the phonological and deep alexia groups (*p* =.06), WNL and phonological alexia groups (*p*= .27) and the deep and global alexia groups (*p*= .87) (See Figure 3).

INSERT FIGURE 3 ABOUT HERE

Within-group comparisons. For the WNL and global alexia groups, paired t-tests revealed no significant within-group differences in semantic and phonologic performance (*p* =.30; *p* = .58, respectively). Within both the phonological alexia and deep alexia groups, semantic abilities were significantly superior to phonologic abilities (t (22) =4.66, p = <.001; t (3) = 3.57, p =.04, respectively) (See Figure 4).

INSERT FIGURE 4 ABOUT HERE

Post-hoc analysis. During visual analysis of the data, individuals with the largest discrepancy between semantic and phonologic abilities (S-P score) appeared to also have the largest lexicality effect, or discrepancy between real word and nonword reading ability (RW-NW score). In order to explore this observation, a simple linear regression model with S-P score predicting RW-NW score was performed with a statistically non-significant outcome (p =.06).

Discussion

Motivated by the PSH, this study investigated how non-orthographic semantic, phonologic, and syntactic abilities inform reading performance in 43 individuals with chronic, stroke-induced aphasia. The current work examined the relationship between non-orthographic language abilities and 1) oral reading abilities, 2) silent reading comprehension abilities, and 3) alexia severity. Results are interpreted per research question before general discussion.

<u>RQ 1: Relationship between non-orthographic language abilities and oral reading</u>

Semantic influence on oral reading. The correlation and regression analyses indicate that non-orthographic semantic abilities are highly related to oral reading abilities in aphasia. Consistent with previous work (Crisp & Lambon Ralph, 2006), the auditory synonym judgment and comprehension of verbs and adjectives tasks showed stronger correlations with oral reading, whereas word-to-picture matching and picture association tasks showed weaker correlations. This pattern suggests picture-based semantic tasks, which primarily tap conceptual-semantic knowledge, are not closely related to oral reading ability. The semantic composite score was significantly predictive of overall oral reading ability (i.e., all word types combined) with the greatest impact on real word reading (irregular > regular) and smaller, yet significant, influences on pseudohomophone and nonword reading.

Given that semantic knowledge is thought to act as a mediating factor for words that have less consistent orthographic-phonologic connections (Ueno et al., 2014), semantic performance was expected to strongly relate to real word reading, particularly irregular words. It was also anticipated that semantics would inform pseudohomophone reading (e.g., hevin), to some degree, since the phonemic sequence of these words corresponds to a real lexical item that has an irregular spelling (e.g., spelling “heaven” as “hevin”). It was initially surprising, however, to find that semantics significantly correlated with and predicted nonword reading since it is commonly thought that nonwords do not benefit from semantic knowledge. This finding upholds the PDP assumption that connections between orthography and semantics cannot be selectively turned off in a parallel-distributed connectionist network, and therefore, nonwords will consequently engage semantic units to some extent (Welbourne & Lambon Ralph, 2007). In agreement with this notion, work with neurologically healthy individuals has shown reading of nonwords is directly influenced by lexical knowledge (Kay & Marcel, 1981; Rosson, 1983). Moreover, Bourassa and Besner (1998) concluded nonwords that resemble real words can effectively prime real words. The nonwords used in the current study all closely resemble real words, and therefore, it is plausible individuals with more intact semantic-orthographic connections may have benefited from the overlap in the orthography of a nonword (e.g., glope) and its parent word (e.g., globe). This can be seen in the number of lexicalization reading errors (e.g., reading a nonword as a real word) made by the participants. From the list of 20 nonword stimuli, an average of 3.44 (*SD* = 4.37; range 0-11) lexicalization errors were made, such as reading “fig” for “flig”, “member” for “merber”, or “dust” for “dusp”. These errors display a lexical bias and suggest that lexical-semantic knowledge was influencing nonword reading performance.

Phonologic influence on oral reading. In agreement with the literature (Crisp & Lambon Ralph, 2006; Henry et al., 2012; Patterson & Marcel, 1992; Rapcsak et al., 2009), the correlation and regression analyses indicate non-orthographic phonologic abilities are highly related to oral reading abilities in individuals with aphasia. However, compared to the semantic correlation results, the order of association was an expected inverse with the strongest phonologic correlations being with pseudohomophones, followed by nonwords, and then real words. It is possible that phonologic performance was more highly correlated with pseudohomophone reading than nonword reading because the phonology of pseudohomophones is more familiar than the unfamiliar phonologic patterns of nonwords that require the language system to form and settle on new patterns of activation (Patterson & Marcel, 1992). Of the four phonologic tasks, the phoneme manipulation task requiring parsing/blending skills showed the strongest correlations with pseudohomophone and nonword reading, which is in agreement with work in both the acquired (Rapcsak et al., 2009) and developmental literature (Bishop & Snowling, 2004; Torgesen, Wagner, & Rashotte, 1994) that has shown individuals with poor phonological awareness and poor phonological short-term memory also tend to have poor decoding abilities.

The regression analyses revealed the phonologic composite score was significantly predictive of total oral reading (i.e., all word types combined) with unique influence on pseudohomophone and nonword reading. It is fitting that phonologic knowledge appears to play a larger role in reading of words with weak or nonexistent semantics; however, it was anticipated phonology would also significantly predict regular word reading given the more predictable relationship between the graphemes and corresponding phonemes of these words. Orthographic-phonologic connections are often disrupted post-stroke (Jefferies et al., 2007) and, therefore, grapheme-phoneme translation may have been avoided, or impossible. Another alternative explanation is that developmentally the reading of familiar, regular real words may rely more on orthographic-semantic connections than orthographic-phonologic connections. Regardless, either explanation would account for a reduced influence of phonologic knowledge on real word reading aloud.

RQ2: Relationship between non-orthographic language abilities and reading comprehension

Semantic influence on silent reading comprehension. Compared to the phonology and syntax composite scores, the semantic composite displayed the strongest relationship with silent reading comprehension. Similar to the oral reading results, the synonym judgment and comprehension of verbs and adjective tasks showed stronger correlations with reading comprehension than the picture-based semantic tasks, further suggesting lexical knowledge, as opposed to conceptual knowledge, has a greater association with reading ability in PWA. As expected, semantics was most predictive of single word reading comprehension indicating that comprehension of spoken words and comprehension of written words is closely linked.

It was unanticipated that the semantic composite would be the greatest predictor of performance on written discourse (i.e., paragraph) comprehension since discourse processing is known to rely on a number of cognitive and linguistic variables (Meteyard, Bruce, Edmundson, & Oakhill, 2015; Peach & Coelho, 2016; Webster, Morris, Howard, & Garraffa, 2018). It is feasible this finding is a reflection of the selected paragraph stimuli. One of the tasks (RCBA 7) involved participants pointing to a picture that corresponded with highly imageable, content words in the paragraph. Therefore, to successfully complete this task, one only needed to match a word or two from the story with one of the picture choices. The other paragraph tasks (RCBA 8 and 9) involved pointing to one of three printed words to complete a sentence. Half of the correct words were found directly in the paragraph, and therefore one only needed to match two words (the word in the story and the word listed as an answer choice) to arrive at the correct answer. Therefore, some participants may have relied solely on orthographic-semantic connections to complete the task, instead of reading the entire passage. In fact, some participants anecdotally reported they were searching for familiar words, which is similar to some of the participants in a study by Knollman-Porter et al. (2015) who reported they opted to skim text for key words they knew they could comprehend.

Syntactic influence on silent reading comprehension. Non-orthographic syntactic performance was significantly related to reading comprehension. The syntactic tasks showed the highest correlations with sentence-level reading comprehension, compared to single word and paragraph reading comprehension. Furthermore, the regression results revealed syntactic performance was uniquely predictive of written sentence comprehension. These findings support the notion that shared syntactic processing supports comprehension of both spoken and written sentences and provides initial evidence that the PSH may extend beyond single word processing.

In addition to sentence-level reading, syntax was anticipated to be uniquely predictive of paragraph comprehension; however this relationship was non-significant. This unexpected result might reflect the nature of the items on the reading measures, as previously mentioned. The isolated sentences comprising the sentence-level reading tests were complex and lacking pragmatic context (e.g., “The dancer is painted by the policeman”; “The carpet the cat is on is green”), which likely engaged syntactic knowledge to a greater degree than the paragraphs that contained simpler, pragmatically contextualized sentences. One could argue that PWA may have been able to recognize individual content words and gather some context when reading the paragraphs, which could have minimized the role of syntax in paragraph reading.

Phonologic influence on silent reading comprehension. The phonologic composite score was found to be significantly correlated with reading comprehension at the single word, sentence, and paragraph level and uniquely predictive at the sentence-level only, per the regression findings. The finding that phonology was significantly predictive of sentence-level, and not word-level, reading comprehension may seem contradictory. However, most sentences contain some words (e.g., functors, abstract words, unfamiliar words) that do not benefit as much from semantic knowledge and, in these cases, it is conceivable that orthographic-phonologic knowledge, in addition to syntactic context, may play a role in decoding and comprehending these words at the sentence-level. An alternate explanation to why phonology is related to sentence-level comprehension is that there is a larger demand on phonological memory to hold the words of a sentence in memory as semantics is activated for comprehending the sentence.

Successful written word comprehension can occur without full access to each phoneme in the target word (Crisp et al, 2011; Jefferies et al., 2007), and this may partially explain why phonology was not significantly predictive at the single word level. Additionally, participants could have been bypassing grapheme-phoneme correspondence and relying more on whole word recognition, as previously mentioned. The degree of relationship between phonology and written word comprehension is likely to change depending on the type of word being read. For example, had written comprehension of only low-frequency words and abstract words, which often have less rich semantic associations, been assessed, phonology may have shown a different, and perhaps stronger, relationship with single word reading comprehension.

<u>RQ 3: Relationship between alexia profile and non-orthographic language performance</u>

Reading profiles. It is known that aphasia and alexia commonly co-occur (Brookshire et al., 2014), and that phonological-deep alexia is the most frequent alexia subtype in aphasia (Luzzatti et al., 2006; Rapcsak et al., 2009). Therefore, it is fitting with the literature that the vast majority of PWA in the current study presented with alexia, and furthermore, 73% (27/37) of the participants with alexia were identified with phonological or deep alexia. Regarding the five PWA whose reading profiles could not be classified, three of these individuals likely presented with a mild phonological alexia evidenced by their small lexicality effects, which precluded a phonological alexia classification; yet, their reading was still outside of normal limits. The other two individuals demonstrated impaired reading and met the deep alexia criteria in terms of reading accuracy (i.e., poor real word and very poor nonword reading), but not in terms of reading errors. Both individuals previously received therapy focused on grapheme-phoneme correspondences and, due to this training, attempted to sound out each word, likely preventing semantic reading errors.

Semantics, phonology, and alexia symptoms. Semantic and phonologic abilities were found to correlate with hallmark alexia symptoms providing another indication that reading performance is linked to non-orthographic language performance. Interestingly, semantic performance was positively correlated with frequency of regularization errors (i.e., errors made from over-reliance on orthographic-phonologic knowledge, such as reading "chef" as "ʃɛf"). This contrary finding might be the result of very few regularization errors being produced. This error type was by far the least common across all participants, with most participants not producing a single regularization error. That is not surprising given regularization errors are associated with surface alexia and no participants fit that reading profile.

As anticipated, phonology was negatively correlated with omission and unrelated reading errors, however, unexpectedly positively correlated with visual/phonologic reading errors. Improved phonology may allow for strengthened, although not perfect, orthographic-phonologic knowledge and therefore more visual/phonologic reading errors are possible when this knowledge is only partially enhanced.

In addition to reading errors, semantic and phonologic abilities have shown similar associations with naming errors in PWA. For example, previous work found semantic abilities to be negatively correlated with semantic errors in naming (Lambon Ralph et al., 2002; Martin, Schwartz, & Kohen, 2006) and reading (Crisp & Lambon Ralph, 2006). Similarly, phonologic abilities were negatively associated with omission errors in naming (Lambon Ralph, Moriarty, & Sage, 2002; Minkina et al., 2016) and reading (current study). These parallel findings across language modalities indicate different language activities (e.g., naming and reading) have similar relationships with semantic and phonologic abilities, supporting the primary systems view of language.

*Semantics, phonology, and alexia profile***.** The data in this study suggest, as others (Crisp & Lambon Ralph, 2006; Leff & Starrfelt) have, that the alexia reading continuum may reflect a semantics-phonology continuum with severity of alexia depending on severity of semantic and phonologic impairment. This is evidenced by the less skilled readers in this sample (i.e., deep and global alexia) all performing below the average semantic and phonologic composite scores, and the more skilled readers (i.e., WNL and phonological alexia) showing average to above average performance. Moreover, the WNL group displayed significantly better semantic and phonologic performance than those in the deep and global alexia groups, and the phonological alexia group demonstrated significantly better semantic and phonologic performance compared to the global alexia group.

In line with the PSH, we expected to find a significant difference in semantic performance between the phonological and deep alexia groups; however this comparison was non-significant (p =.06). This finding might reflect the difficulty of precisely characterizing an individual as having phonological or deep alexia given the overlapping symptoms and the fact that people can evolve from a deep to phonological alexia profile (Friedman, 1996; Leff & Starrfelt, 2014). A lack of significant difference (p =.27) in phonological performance between the WNL and phonological alexia groups was also unexpected. This result might be related to the possibility that the “normal” readers are not, in reality, “normal” since they were identified based only on oral reading of single words and might display reading impairment beyond the word-level.

Within group comparisons revealed the phonological and deep alexia groups demonstrated superior semantic, compared to phonologic, performance, supporting PSH predictions. Furthermore, a post-hoc analysis showed the discrepancy between semantic and phonologic ability (S-P score) approached a significant, positive relationship with the lexicality effect (RW-NW score) hinting that this hallmark feature of phonological-deep alexia might represent more than an imbalance of real word and nonword reading.

Overall, these findings align with the PSH and imply that a person's non-orthographic semantic and phonologic abilities underpin his/her reading profile. Specifically, the data suggest a PWA who demonstrates relatively intact semantic and phonological abilities is likely to display mild reading difficulties, whereas, an individual with superior semantic versus phonologic abilities is likely to fit a phonological-deep alexia profile, and an individual with severely impaired semantics and phonology is expected to have a reading profile consistent with global alexia.

General Discussion

Viewed collectively, results from this study provide support for the PSH view of language processing that proposes spoken and written language impairment are strongly related. This study confirms findings from previous work (Crisp & Lambon Ralph, 2006; Henry et al., 2012) that showed non-orthographic semantic and phonologic abilities are predictive of oral reading abilities in individuals with aphasia (RQ1). In addition, this work provides evidence that the PSH framework may extend beyond oral reading of single words by demonstrating non-orthographic language abilities are also significantly related to silent reading comprehension at the word, sentence, and paragraph levels (RQ 2). Finally, the data suggests alexia may reflect underlying central language impairment with severity of non-orthographic semantic and phonologic impairment corresponding to severity of alexia (RQ 3).

If applied to clinical practice, these findings indicate the importance of routine assessment of reading abilities and integrated treatment of written and spoken language impairments in aphasia. Results from this study endorse recent alexia/agraphia treatments (e.g., Beeson et al., 2010; Beeson, Rising, DeMarco, Foley, & Rapcsak, 2018; Brookshire et al., 2014; Johnson, Ross, & Kiran, 2017) that, in addition to orthographic ability, also assessed and treated non-orthographic phonology and/or semantics in an effort to improve written language processing. Additionally, findings from this study encourage use of performance on non-orthographic semantic and phonologic tasks to aid in the diagnosis of alexia instead of relying solely on single word oral reading accuracy and types of oral reading errors produced, as is commonly practiced.

This work is preliminary and needs to be replicated and extended to further understand the relationship between acquired spoken and written language impairment in aphasia. Future research can address limitations of the current study in a number of ways. First, participant inclusion criteria can be expanded to include both acute and chronic aphasia, as well as aphasia due to etiologies other than stroke. Moreover, each person's lesion location could be correlated with reading performance to better relate type of aphasia and alexia. Second, the selection of language measure can be more diverse. Specifically, in addition to the receptive, non-orthographic language tasks utilized in this study, spoken tasks should also be included in the composite language scores so the influence of expressive semantic, phonologic, and syntactic abilities on oral reading and silent reading comprehension can be explored. The impact of other predictors, such as attention and memory, on written language ability should also be addressed.

The influence of other psycholinguistic variables, such as degree of word imageability, concreteness, and part of speech (especially inclusion of function words and bound morphemes), on reading performance should be examined. More advanced reading stimuli, such as oral reading of text and comprehension of more complex written paragraphs should be considered, as well. Reading speed, in addition to accuracy, can also be measured to better capture functional reading performance. Furthermore, to fully assess the relationship between acquired spoken and written language impairments, spelling at the word and sentence-level should be included, in addition to reading tasks. Finally, it would be particularly interesting and informative to examine the PSH in a non-alphabetic orthographic system to determine whether the PSH predictions can be supported cross-linguistically and to a different type of orthographic system.

In conclusion, we hope that these findings encourage researchers and clinicians to continue to investigate the nature of acquired written language deficits, and importantly, to routinely examine both spoken and written language abilities in individuals with aphasia. This practice has the potential to identify commonalities between the two that may be amenable to therapeutic intervention.

Acknowledgements

This work was supported by the American Speech-Language-Hearing Foundation (ASHF) New Century Scholars Doctoral Scholarship and the University of Washington Training Grant in Speech and Hearing Sciences (T32000033). The authors would like to thank Lauren Belongia, Shanni Gellar, Nicole Okada, and Alexa Wilkenson for their assistance on this project, and the dedicated participants with aphasia who volunteered to take part in this study.

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Tables

Table 1. Assessment battery

Language Domain	*Type of Assessment*	*Task (Number of items)*
Semantics	(Non-orthographic) Conceptual and lexical knowledge	CCT: Picture Association (64)
		PALPA 47: Spoken word-to-picture match (40)
		PALPA 49: Auditory synonym judgment (60)
		PALPA 57: Auditory comprehension of verbs & adjectives (41)
Phonology	(Non-orthographic) Phonological awareness	SAPA: Minimal pairs (13)
		SAPA: Real word rhyme judgment (15)
		SAPA: Nonword rhyme judgment (22)
		LAC: Parsing/blending sounds in isolation (16) and syllables (12)
Syntax	(Non-orthographic) Syntactic processing	CAT 9: Comprehension of spoken sentences (16)
		PALPA 55: Spoken sentence-to-picture match (60)
		PALPA 58: Auditory comprehension of locative relations (24)
Oral Reading	Single words	ABRS regularly spelled words (e.g., broom) (40)
		ABRS irregularly spelled words (e.g. yacht) (40)
		ABRS nonwords (e.g., flig) (20)
		SAPA pseudohomophones (e.g., hevin) (12)
Silent Reading Comprehension	Single words	CAT 8: Written word-to-picture match (15)
		PALPA 50: Written synonym judgment (60)
		PALPA 51: Written word semantic association (30)
	Sentences	CAT 10: Written sentence-to-picture match (16)
		PALPA 59: Written comprehension of locatives (24)
		RCBA 4: Functional Sentence Reading (10)
	Paragraphs	RCBA 7: Paragraph-to-picture match (10)
		RCBA 8: Paragraph-factual (10)
		RCBA 9: Paragraph-inferential (10)

CCT = Cactus and Camel Test (Adlam et al., 2010); PALPA = Psycholinguistic Assessment of Language Processing in Aphasia (Kay et al., 1992); SAPA = Standardized Assessment of Phonology in Aphasia (Kendall et al., 2010); CAT =Comprehensive Aphasia Test (Swinburn et al., 2004); LAC = Lindamood Auditory Conceptualization Test (Lindamood & Lindamood, 1988); ABRS = Arizona Battery of Reading and Spelling

Table 2. Performance on non-orthographic language and silent reading comprehension tasks (n=43)

	Non-orthographic Language		**Oral Reading**					
	Semantic Composite	*Phonology Composite*	*Regular Words*	*Irregular Words*	*All Real Words*	*Non-words*	*Pseudo-homophones*	*Composite (all words)*
AVG	83%	76%	74%	69%	72%	40%	45%	63%
SD	10%	10%	33%	34%	33%	35%	33%	31%
Range	55-98%	53-97%	3-100%	3-100%	3-100%	0-100%	0-100%	2-97%

Table 3. Multiple linear regression results with semantics and phonology as predictors of oral reading

				Standard Regression			
	R^2_{total}	R^2_{Adj}	F_{total}	b	*(SE)*	t	p-value
Total Oral Reading	*.57*	*.54*	*26.07(2,40)****				
Semantics				0.54	(0.14)	3.70	0.00 **
Phonology				0.44	(0.16)	2.71	0.01 *
Regular Word Reading	*.49*	*.46*	*18.98(2,40)****				
Semantics				0.64	(0.17)	3.78	0.00 **
Phonology				0.31	(0.19)	1.60	0.12
Irregular Word Reading	*.53*	*.51*	*22.64(2,40)****				
Semantics				0.71	(0.16)	4.38	0.00 ***
Phonology				0.26	(0.18)	1.43	0.16
Pseudohomophone Reading	*.57*	*.55*	*26.28(2,40)****				
Semantics				0.38	(0.16)	2.26	0.02 *
Phonology				0.69	(0.18)	3.95	0.00 ***
Nonword Reading	*.40*	*.37*	*13.24(2,40)****				
Semantics				0.40	(0.18)	2.18	0.04 *
Phonology				0.50	(0.21)	2.40	0.02 *

* $p < .05$, ** $p < .01$, *** $p < .001$.

Table 4. Performance on non-orthographic language and silent reading comprehension tasks (n=43)

	Non-orthographic Language			**Silent Reading Comprehension**			
	Semantic Composite	*Phonologic Composite*	*Syntax Composite*	*Single Words*	*Sentences*	*Paragraphs*	*Composite (all levels)*
AVG	83%	76%	67%	79%	69%	81%	76%
SD	10%	10%	19%	12%	20%	20%	17%
Range	55-98%	53-97%	37-97%	40-95%	19-100%	13-100%	24-96%

Table 5. Multiple linear regression results with semantics, phonology, and syntax as predictors of silent reading comprehension

				Standard Regression			
	R^2_{total}	R^2_{Adj}	F_{total}	*b*	*(SE)*	*t*	*p*-value
Total Reading Comprehension	*.72*	*.70*	*33.03(3,39)****				
Semantics				0.55	(0.14)	3.85	0.00 ***
Phonology				0.22	(0.13)	1.69	0.10
Syntax				0.19	(0.13)	1.46	0.15
Single Words	*.63*	*.60*	*21.98(3,39)****				
Semantics				0.71	(0.17)	4.14	0.00 ***
Phonology				0.13	(0.15)	0.89	0.41
Syntax				0.07	(0.16)	0.45	0.66
Sentence Level	*.81*	*.79*	*54.25(3,39)****				
Semantics				0.39	(0.12)	3.20	0.003 **
Phonology				0.23	(0.11)	2.11	0.04 *
Syntax				0.41	(0.11)	3.65	0.00 **
Paragraph Level	*.57*	*.54*	*17.16(3,39)**				
Semantics				0.54	(0.19)	2.89	0.006 **
Phonology				0.30	(0.17)	1.72	0.09
Syntax				0.10	(0.17)	0.56	0.57

* $p < .05$, ** $p < .01$, *** $p < .001$ (two-tailed).

Table 6. Non-orthographic semantic and phonologic performance per reading profile

Reading Profile	***Avg. Semantic Composite***		***Avg. Phonologic Composite***	
	%	z-score	%	z-score
WNL	88% (5%)	0.39 (0.42)	82% (11%)	0.35 (0.79)
Phonological Alexia	85% (10%)	0.2 (0.83)	77% (11%)	0.11 (0.74)
Deep Alexia	75% (3%)	-0.61 (0.36)	67% (6%)	-0.57 (0.56)
Global Alexia	68% (8%)	-1.17 (0.68)	66% (7%)	-0.75 (0.48)

Standard deviation shown in parenthesis; WNL = within normal reading limits; Avg. = average

Figure 1. Schematic of primary systems/connectionist language framework. Note: From "Selective disorders of reading?" by K. Patterson and M. Lambon Ralph, 1999, *Current Opinion in Neurobiology*, 9, p. 235. Copyright 1999 by Elsevier Science Ltd. Reprinted with permission.

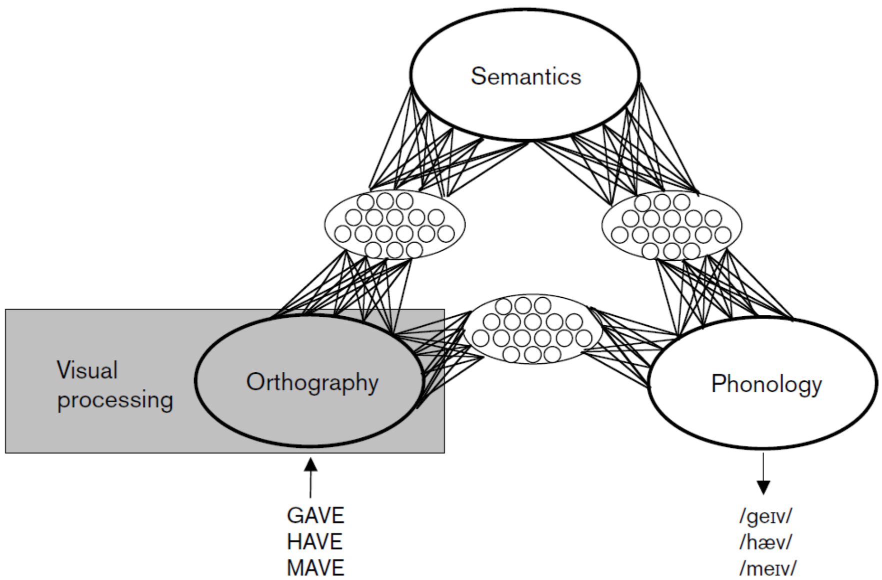


Figure 2. Proposed relationships between degree of reading impairment (alexia type) and degree of phonological and semantic impairment. Note: From “Unlocking the nature of the phonological-deep dyslexia continuum: The keys to reading aloud are in phonology and semantics” by J. Crisp and M. Lambon Ralph, 2006, *Journal of Cognitive Neuroscience*, 18, p. 348. Copyright 2006 by The MIT Press. Reprinted with permission.

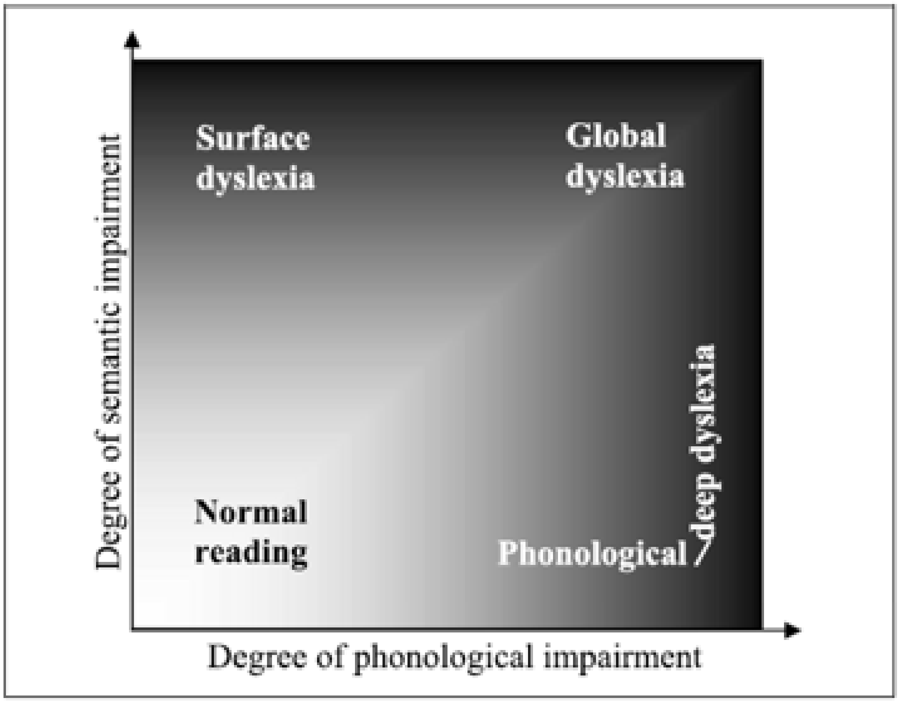
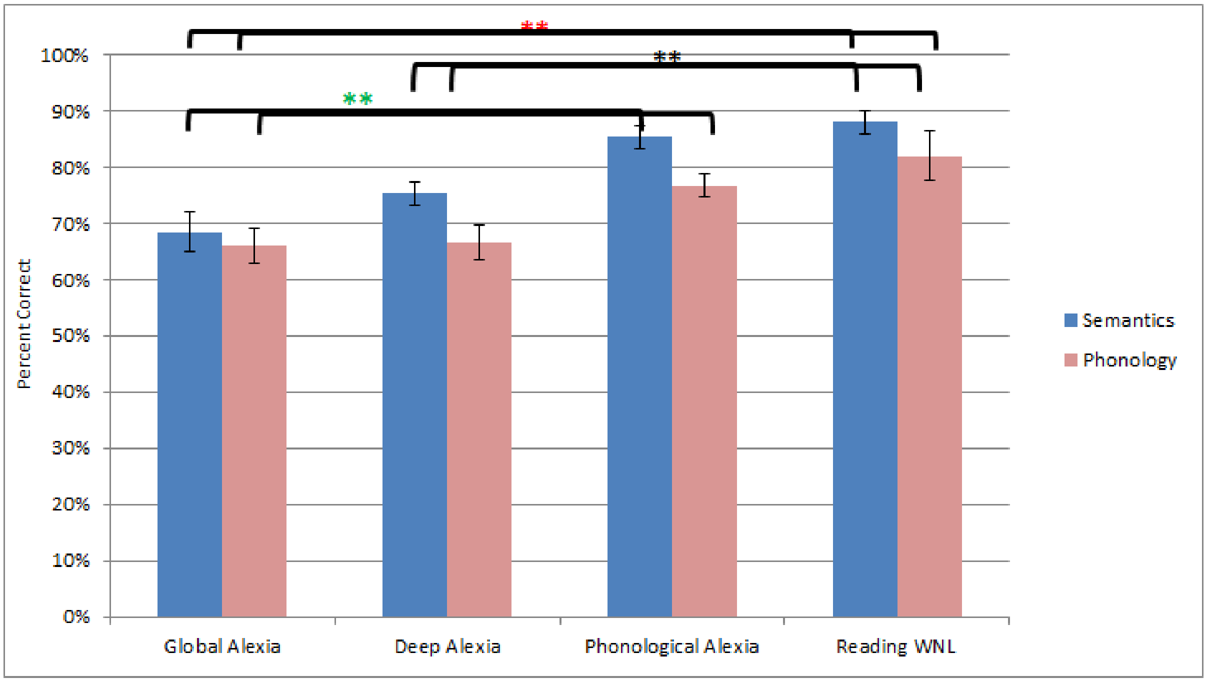
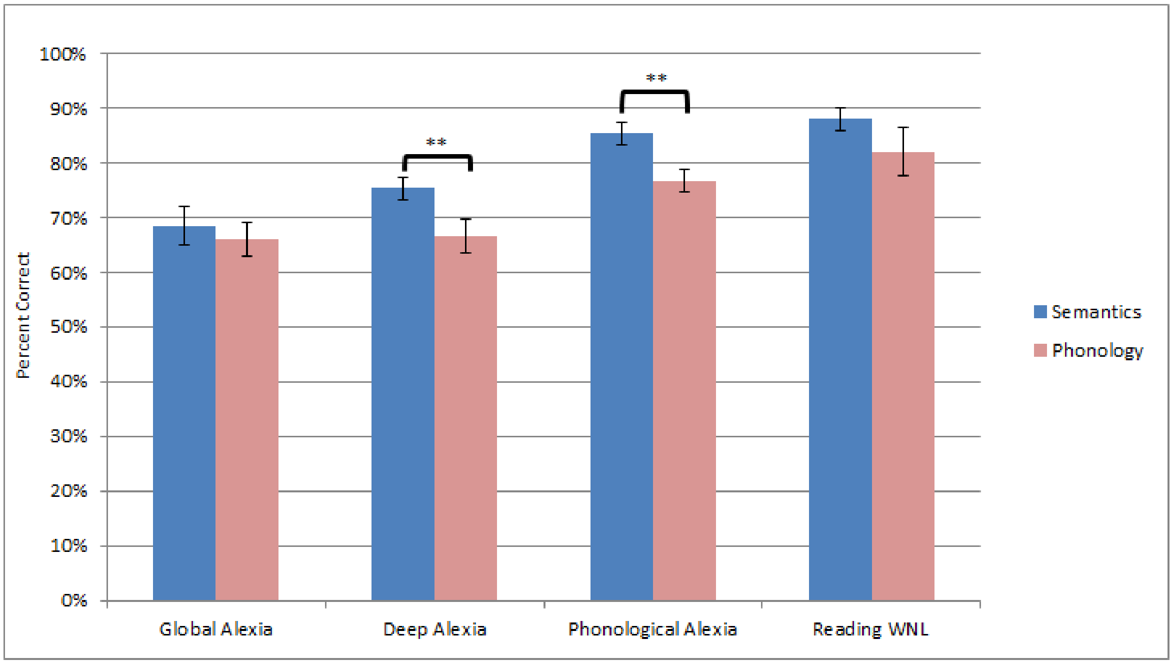


Figure 3. Between–group differences in non-orthographic semantic and phonologic performance



** = difference between WNL and global; ** = difference between WNL and deep; ** = difference between phonological and global; error bars represent standard error

Figure 4. Within–group differences in non-orthographic semantic and phonologic performance



** = statistically significant within-group difference; error bars represent standard error

Supplementary Appendix A

ID	***Age***	***MPO***	***Education***	***Sex***	***Handedness***	**CAT Spoken Comprehension** (cut off 56)	**CAT Naming** (cut off 69)	**CAT Repetition** (cut off 48*)	**CAT Picture Description** (cut off 33)
P1	55	57	14	F	R	44	57	40	11
P2	77	47	13	F	R	59	69	44	31.5
P3	31	71	14	M	R	40	58	46	18.5
P4	62	82	16	M	R	56	72	42	29
P5	71	112	14	M	L	51	35	28	11.5
P6	71	216	14	F	R	44	58	34	21.5
P7	64	135	16	F	R	40	56	35	22
P8	70	60	18	M	R	56	49	39	19.5
P9	58	131	18	M	L	41	42	34	10
P10	67	65	16	M	R	43	1	20	4
P11	55	79	16	M	R	51	67	44	30
P12	45	16	16	F	R	56	63	48	21
P13	61	76	14	F	R	42	46	32	17.5
P14	51	74	12	F	R	56	58	48	18.5
P15	68	102	22	M	R	49	22	32	15
P16	71	59	20	F	R	57	76	46	39
P17	47	28	17	F	R	57	61	30	23
P18	72	156	16	M	R	54	61	39	20
P19	64	59	17	F	R	58	77	42	28
P20	73	38	18	F	R	60	67	40	30
P21	59	118	21	F	R	43	38	48	12
P22	71	91	12	F	R	32	10	28	7.5
P23	92	18	18	F	R	29	15	6	14.5
P24	65	48	17	F	R	58	57	46	29.5
P25	77	132	18	M	R	43	2	8	7.5
P26	66	109	16	M	R	36	16	0	-3
P27	58	28	12	M	R	55	55	22	21
P29	47	91	13	M	L	40	21	44	3.5
P30	42	139	18	M	R	46	60	23	36.5
P33	68	46	12	M	R	61	79	48	47
P34	63	50	13	M	R	55	39	40	23
P35	56	22	15	M	R	64	72	42	24
P36	66	11	16	M	R	56	60	46	16
P37	62	8	16	M	R	55	56	42	31
P38	73	11	14	F	R	30	15	23	0.5
P39	69	46	12	M	R	51	45	34	10
P41	57	51	12	M	R	42	16	16	3

P42	71	32	20	M	R	47	26	36	6.5
P43	38	47	12	F	R	54	50	48	17
P44	58	96	18	F	L	32	17	12	6
P45	55	26	16	M	R	49	20	46	24.5
P46	66	22	18	M	R	54	77	46	27.5
P47	59	69	16	M	R	48	68	48	17.5
				25 M, 18 F	39 R, 4 L				
AVG	62.12	69.16	15.72			48.70	46.72	35.23	18.67
SD	11.39	45.42	2.62			9.00	22.68	12.72	10.94
Range	31-92	8-216	12-22			29-64	1-79	0-48	-3 - 47

Note: MPO = months post-stroke onset; R = right-handed; L = left-handed; CAT = Comprehensive Aphasia Test (Swinburn et al., 2004); P16 and P33 scored above cut-off scores on 3/4 subtests; however, they were included due to demonstration of marked language difficulties during conversational speech and experimental testing.

*Only repetition of words and nonwords (CAT subtests 12-14) was administered.

Supplementary Appendix B

Semantic Tasks

	CCT	PALPA 47	PALP49	PALPA57
CCT	--			
PALPA 47	.51**	--		
PALPA 49	.68**	.67**	--	
PALPA 57	.45**	.57**	.71**	--

CCT = Camel and Cactus Test (Adlam et al., 2010); PALPA = Psycholinguistic Assessment of Language Processing in Aphasia (Kay et al., 1992)

* *p* < .05, ** *p* < .01, *** *p* < .001.

Phonologic Tasks

	SAPA min pairs	SAPA RW rhyme	SAPA NW rhyme	LAC
SAPA min pairs	--			
SAPA RW rhyme	.41**	--		
SAPA NW rhyme	.35*	.74**	--	
LAC	.26	.36*	.33*	--

SAPA = Standardized Assessment of Phonoloy in Aphasia (Kendall et al., 2010); LAC = Lindamood Auditory Conceptualization Test (Lindamood & Lindamood, 1988)

* *p* < .05, ** *p* < .01, *** *p* < .001.

Syntactic Tasks

	PALPA55	PALPA58	CAT 9
PALPA 55	--		
PALPA58	.76**	--	
CAT 9	.85**	.71**	--

PALPA = Psycholinguistic Assessment of Language Processing in Aphasia (Kay et al., 1992); CAT = Comprehensive Aphasia Test (Swinburn et al., 2004)

** p < .05, ** p < .01, *** p < .001.*

Oral Reading Tasks

	ABRS regular	ABRS irregular	ABRS nonwords	SAPA pseudo
ABRS regular	--			
ABRS irregular	.98**	--		
ABRS nonwords	.69**	.70**	--	
SAPA pseudo	.78**	.81**	.87**	--

ABRS = Arizona Battery for Reading and Spelling (Beeson et al., 2010); SAPA = Standardized Assessment of Phonoloy in Aphasia(Kendall et al., 2010)

* *p* < .05, ** *p* < .01, *** *p* < .001.

Silent Reading Comprehension Tasks

	Single Words			Sentences			Paragraphs		
	CAT 8	PALPA 50	PALPA 51	CAT 10	PALPA 59	RCBA 4	RCBA 7	RCBA 8	RCBA 9
CAT 8	--								
PALPA 50	.64**	--							
PALPA 51	.63**	.81**	--						
CAT 10	.67**	.75**	.66**	--					
PALPA 59	.69**	.62**	.60**	.78**	--				
RCBA 4	.60**	.67**	.75**	.55**	.54**	--			
RCBA 7	.59**	.59**	.62**	.62**	.71**	.57**	--		
RCBA 8	.64**	.78**	.82**	.66**	.60**	.73**	.64**	--	
RCBA 9	.68**	.76**	.75**	.75**	.72**	.76**	.68**	.87**	--

CAT = Comprehensive Aphasia Test (Swinburn et al., 2004); PALPA = Psycholinguistic Assessment of Language Processing in Aphasia (Kay et al., 1992)RCBA = Reading Comprehension Battery for Aphasia (LaPointe & Horner, 1982)

* *p* < .05, ** *p* < .01, *** *p* < .001.