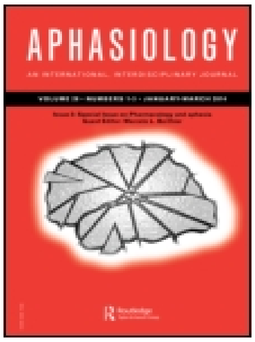

Aphasiology

ISSN: 0268-7038 (Print) 1464-5041 (Online) Journal homepage: http://www.tandfonline.com/loi/paph20

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Aphasiology, 2014
Vol. 28, No. 12, 1464–1480, http://dx.doi.org/10.1080/02687038.2014.945389

Frequency, nature, and predictors of alexia in a convenience sample of individuals with chronic aphasia

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Background: It is broadly known that persons with chronic aphasia experience difficulty reading. However, the frequency of acquired reading disorders (alexia), and the most common subtype of alexia, as well as predictors of reading in this population are yet to be determined.
Aims: This study aims to provide initial evidence regarding the frequency, nature, and predictors of alexia in a large convenience sample of persons with chronic aphasia.
Methods & Procedures: Single-word oral reading abilities for lexical items (regular and irregular words) and sublexical items (pseudohomophones and nonwords) from 99 persons with aphasia (PWA) and 29 normal controls (NC) were assessed and retrospectively analysed.
Outcomes & Results: Of the 99 PWA, 68% met our alexia criteria. These PWA and coexisting alexia performed worse than the NC on all reading stimuli and tended to perform worse with stimuli requiring sublexical processing (i.e., pseudohomophones and nonwords) than on stimuli requiring lexical processing (i.e., real words). The group of PWA and alexia had a wide range of aphasia types and severities. Less severe aphasia was found to predict higher oral reading performance. Education was not found to be a significant predictor of reading.
Conclusions: Our retrospectively analysed results from a convenience sample suggest that reading problems occur frequently among PWA and severity of aphasia influences reading performance. Moreover, our results suggest that acquired reading difficulties after stroke are likely to be characterised by difficulty with sublexical processing. A

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The authors would like to thank all participants as well as members of the University of Washington Aphasia Research Laboratory, particularly Amanda Hendricks, for their contributions to this project. This study was supported by the VA RR&D Brain Rehabilitation and Research Center, VA RR&D [grant number C3759K] and the University of Washington Aphasia Research Laboratory Registry and Repository Database.

priori work is needed to provide greater control over participant and stimuli selection to further systematically explore the presence and nature of alexia within chronic aphasia.

Keywords: Alexia; Aphasia; Reading; Frequency; Predictors.

Persons with aphasia (PWA) often have acquired reading difficulties (alexia); however, little is known about the frequency, subtypes, and predictors of aphasia-associated alexia. In a convenience sample of 99 PWA, we sought to quantify and qualify coexistent alexia utilising a two-route (sublexical and lexical) reading classification model.

Reading aloud is thought to occur by two routes: (1) a sublexical route whereby orthographic sequence representations are mapped directly onto phonological sequence representations and (2) a lexical, or whole word, route by which orthographic representations are mapped onto semantic representations, which are then mapped onto phonological representations (Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001; Coslett, 2011). From a computational connectionist model perspective (e.g., Plaut, 1999; Plaut, McClelland, Seidenberg, & Patterson, 1996; Seidenberg & McClelland, 1989) the sublexical route, in the course of its experience with language, has acquired knowledge of the orthographic-phonologic sequence regularities of the language, which are encoded in neural connectivity. These learned statistical regularities provide the basis for implicit "grapheme-phoneme correspondence" rules. Both regular and irregular words, however infrequent, can take advantage of these implicit rules, regular words to the greatest degree, but also irregular words (e.g., laugh) that share a sequence relationship with a number of other irregular words. These implicit, neurally instantiated rules provide the only basis for reading nonwords (words that do not violate English grapheme to phoneme correspondence rules yet lack meaning). Damage to this sublexical route is associated with reading errors involving phonologic sequence.

The lexical route also supports the reading of both regular and irregular words. However, because it is substantially a whole word route, it cannot instantiate much knowledge of regularities in the relationships between orthography and phonology. Thus, frequency and age of acquisition, both reflected in the robustness of instantiation in neural connectivity, are the dominant factors in determining resistance to network damage or noise. Nonwords cannot be processed by this lexical pathway. Given that this pathway incorporates the substrate for semantics, damage to it may, with some lesions, lead to production of semantic paralexias.

This conceptualisation of lexical and sublexical reading routes has been used previously to classify central alexia into three main subtypes (i.e., surface, phonological, and deep).

These subtypes are typically differentiated on the basis of the pattern of errors (paralexias) made during oral reading of regular words (e.g., dog), irregular words (e.g., yacht), pseudohomophones (e.g., fyte), and nonwords (e.g., dessy) (Cherney, 2004). Surface alexia stems from damage to the lexical reading route. It is associated with difficulty reading irregularly spelled words, particularly low-frequency (LF) words, with relatively preserved reading of regularly spelled words and nonwords. In contrast, phonological alexia stems from damage to the sublexical reading route. It is characterised by impaired reading of nonwords and unfamiliar, often LF, words.

Deep alexia resembles phonological alexia; however, due to additional partial damage to the lexical route, semantic errors may occur in addition to phonologic errors for both real words and nonwords (Coslett, 2000; Plaut et al., 1996). Given that both phonological and deep alexia involve sublexical route damage and often have overlapping errors (except semantic errors), these two subtypes have been viewed as points along an impairment continuum (Crisp & Lambon-Ralph, 2006; Friedman, 1996). These findings inform our approach to alexia classification, and two distinct forms of reading disability are highlighted in our study: lexical (surface alexia) and sublexical (phonological and deep alexia).

While it is broadly known that alexia can occur in the presence of aphasia (Beeson & Hillis, 2001; Cherney, 2004; Riley & Kendall, 2013; Webb & Love, 1983), the frequency of aphasia-associated alexia, its subtypes, and the nature of predictors are uncertain, even as these factors may have implications for aphasia rehabilitation programmes. Most studies of alexia in aphasia have been focused on treatment and, as a result, have been restricted to small sample sizes (Cherney, 2004; Coslett, 2000). These smaller sample sizes limit the ability to provide widely generalisable information about the frequency of alexia in the larger population of PWA.

In a previous study, we found that 80% of 41 people with a wide range of aphasia types presented with alexia and that phonological alexia was the most common subtype of central alexia (Wilson, Gonzalez Rothi, Nadeau, & Kendall, 2007). Furthermore, in a group of 35 PWA, Webb and Love (1983) found that individuals who had less than high school education read at the single-word and sentence levels significantly worse than those with post-high-school education, and that "overall language disorder" was the best indicator of reading performance.

The present investigation expands upon this prior work (Webb & Love, 1983; Wilson et al., 2007) by retrospectively analysing data from a large convenience sample of PWA to further address questions concerning the frequency, subtypes, and previously reported predictors (i.e., education and aphasia severity) of aphasia-associated alexia.

METHOD

Participants

One hundred persons with aphasia (PWA) and 29 normal controls (NC) were recruited to participate in the standardisation of the Standardised Assessment of Phonology in Aphasia (SAPA; Kendall et al., 2010). The reading data collected from Subtest 1 of the SAPA (described later) were retrospectively analysed for this study.

Forty-eight PWA and 29 NC were recruited through the Department of Veterans Affairs Rehabilitation Research and Development Brain Rehabilitation Research Center in Gainesville, Florida, and consented according to an Institutional Review Board (IRB) approved protocol. Data from 41 of these PWA were also reported in the Wilson et al. (2007) study. An additional 52 PWA were recruited through the University of Washington (UW) Aphasia Research Laboratory Registry and Repository Database and consented under a separately approved IRB protocol.

The inclusion criteria for PWA were history of left hemisphere stroke at least 6 months prior to enrolment and a diagnosis of aphasia confirmed by a licenced speech–language pathologist based on an aphasia quotient (AQ) of less than 93.8/

100 on the Western Aphasia Battery (WAB) (Kertesz, 1982) and performance below 56/60 on the Boston Naming Test (BNT; Kaplan, Goodglass, Weintraub, & Segal, 1983).

To confirm that NC participants did not have evidence of impaired cognitive or linguistic abilities, the Mini-Mental Status Examination (Folstein, Folstein, & McHugh, 1975), the BNT (Kaplan et al., 1983), and the National Adult Reading Test (Nelson, 1982) were administered. These measures are widely used to assess cognitive status, picture naming abilities, and irregular word reading abilities, respectively. All control participants performed within normal limits on each of these measures.

Exclusion criteria for both PWA and NC groups included preexisting neurological disease and/or uncorrected moderate to severe vision or hearing impairment. Amount of education was not an a priori inclusion/exclusion criterion; however, one PWA who had only 1 year of formal education was excluded from subsequent analysis to obviate potential skewing of the results. Thus, our final sample included 99 of the 100 PWA consented for the study.

Stimuli

The stimuli were created as part of the SAPA, a comprehensive test of phonologic processing that assesses (1) oral reading, (2) auditory phonologic processing, and (3) repetition, parsing, and blending of phonologic sequences. These three subtests were conceptualised to measure a construct of phonology. The stimuli employed in this study make up only the items on Subtest 1, which assesses oral reading abilities for regularly and irregularly spelled words, pseudohomophones, and nonwords. Regular and irregular words can be read by the lexical and/or sublexical route, whereas pseudohomophones and nonwords rely on the sublexical reading route.

The regularly spelled words (n = 21) were words whose spelling followed common grapheme–phoneme correspondences (e.g., job), whereas the irregularly spelled words (n = 11) violated these typical letter–sound patterns in English (e.g., laugh). The pseudohomophones (n = 8) were composed of unfamiliar orthography with familiar phonology and meaning (e.g., fyte, pronounced fight). Pseudohomophones were created using one-syllable "parent" nouns that were converted into a pseudohomophone by the exchange or addition of phonemically consistent or neutral graphemes (e.g., fight became fyte). The nonwords (n = 12) did not violate English grapheme to phoneme correspondence rules and lacked meaning (e.g., dessy). The nonwords were created to represent four-syllable patterns ranging from simple to more complex (i.e., CVC, CVCV, CVCVC, and CVCVCVC) and were not created from "parent" real words like the pseudohomophones. See Table 1 for a complete list of the reading stimuli.

Word frequencies listed in Table 2 for the regular, irregular, and pseudohomophone "parent" words reflect values from the Thorndike and Lorge (1944) written frequency corpus, which were obtained from the MRC Psycholinguistic Database (Wilson, 1988). Thorndike–Lorge values were used because all of the reading stimuli on the SAPA could be found in this large corpus of 18,000,000 words. Independent samples two-tailed t-tests revealed no significant difference in average frequency between the regular and irregular words, p = .85, between the regular words and pseudohomophone "parent" words, p = .76, or between the irregular words and pseudohomophone "parent" words, p = .91.

TABLE 1
Oral reading stimuli from Subtest 1 of Standardised Assessment of Phonology in Aphasia (SAPA)

Read via lexical and/or sublexical processing		*Read via sublexical processing*	
Regularly spelled words	*Irregularly spelled words*	*Pseudohomophones*	*Nonwords*
lot	laugh	fyte	nis
job	suit	rhed	kes
red	pew	phine	nush
bin	coup	kupp	dessy
fib	heir	clinck	simite
itch	office	jirm	leedle
baby	promise	rhigg	junooge
city	service	troall	choithane
melon	bodice		sedeatin
vigil	ratio		nysimin
afternoon	rhapsody		shoinaejouth
president			jounaethawn
family			
caramel			
promoter			
meteorite			
diplomacy			
regulator			
disability			
imbecility			
generosity			

TABLE 2
Across word type comparisons for word frequency, length, and part of speech

Word type	*N*	*Frequency mean (SD)**	*Letters mean (SD)*	*Syllables mean (SD)*	*Phonemes mean (SD)*	*Part of speech*
Regular	21	528.81 (697.18)	6.33 (2.78)	2.62 (1.40)	6.1 (2.70)	20 nouns, 1 adjective
Irregular	11	578.55 (695.13)	5.36 (1.57)	1.72 (0.79)	4.1 (1.76)	11 nouns
Pseudohomophone	8	615.25 (646.88)	4.75 (0.89)	1.00 (0.00)	3.38 (0.74)	8 parent nouns
Nonword	12	N/A	6.83 (2.89)	2.08 (0.79)	5 (1.65)	N/A

*Thorndike–Lorge written frequency text corpus values (Thorndike & Lorge, 1944).
Nonword frequencies could not be calculated because the nonwords were not derived from a real word. Instead, high and low phonemic frequency values for consonants and vowels were controlled within the nonwords using Shriberg and Kent (1982) phonemic frequency values.

Nonword frequencies could not be calculated because the nonwords were not derived from a real word. Instead, high and low phonemic frequency values for consonants and vowels were controlled within the nonwords using Shriberg and Kent (1982) phonemic frequency values. Vowel frequency ranges were high frequency (HF) > 7.0 and LF < 4.0, and consonant frequency ranges were HF > 5.0 and LF < 2.0. The Phonetic Probability Calculator (Vitevitch & Luce, 2004) was used to calculate the sum of all biphone probabilities (i.e., probability of sound segments cooccurring within a word) within each nonword. The average sum biphone probabilities were equal across the four nonword syllable patterns.

Concerning word length, the regular, irregular, and nonwords did not significantly differ from one another in average number of letters, syllables, or phonemes. After Bonferroni correction for Type I error (yielding a threshold p value of .01), independent samples two-tailed t-tests revealed no significant difference in average letters, syllables, or phonemes between the regular and irregular words, $p = .22$, $p = .03$, $p = .02$ (respectively), between the regular words and nonwords, $p = .63$, $p = .17$, $p = .16$ (respectively) or between the irregular words and nonwords, $p = .14$, $p = .30$, $p = .27$ (respectively). In contrast, the pseudohomophones significantly differed from the other word types in word length because only one-syllable pseudohomophones were assessed. Pseudohomophones differed from regular words and nonwords in syllables, $p < .001$, $p = .001$ (respectively) and sounds, $p < .001$, $p = .009$ (respectively), and pseudohomophones differed from irregular words only in number of syllables, $p = .01$.

Concerning part of speech, all but one (98%) of the regular, irregular, and pseudohomophone "parent" words were nouns, and therefore, the stimuli are similar regarding word class. Nonwords could not be assigned a part of speech due to lack of "parent" real words.

Data collection and scoring procedure

The orthographic stimuli were presented to participants via Microsoft PowerPoint software on a 20-inch computer monitor. Participants were tested on a single category at a time (i.e., regular words, nonwords, pseudohomophones, and irregular words). Before the presentation of regular and irregular words, the participants saw and heard the following: "You will see a word on the screen. Read the word out loud as best you can. Take your time". Prior to the pseudohomophone and nonword words, the participants saw and heard the instructions: "You will see a made up word on the screen. This word does not mean anything. Read the word out loud as best you can. Take your time". The words were presented in black, bold, lowercase, Arial 72-point font on a white background. The words were shown one at a time, centred on the screen and remained in sight until the participant had completed his or her response. To ensure that participants understood the task, three practice trials of each stimulus type were completed before the test items were presented. Feedback on performance was provided during practice trials only.

Participants' responses were scored by a certified speech–language pathologist or trained research assistant as correct or incorrect (i.e., 1 = correct and 0 = incorrect) according to operational criteria described later, and a portion of participant oral reading trials was digitally recorded for reliability analyses. Verbal productions were scored as correct if the response matched the target stimulus or if the participant produced a close approximation, such as an identifiable phoneme distortion. For nonwords with two possible pronunciations based on English grapheme–phoneme correspondences (e.g., nush), either pronunciation was scored as correct. Nonresponses, semantic errors, and phonologic errors were all scored as incorrect. UW participant responses were recorded as correct or incorrect online, and the test administrator manually advanced the computer screen, while University of Florida (UF) participant responses were scored offline, and a uniform interstimulus interval of 8 s was employed to advance the screen. There was no significant difference in reading performance on the SAPA, $t(97) = -.55$, $p = .58$, naming performance on the BNT, $t(97) = .12$, $p = .91$, and language performance on the WAB-AQ, $t(97) = 1.03$, $p = .31$, between PWA seen at UF and those seen at UW. Due to the retrospective nature of this study, only the number of correct and incorrect responses (as opposed to participants' actual verbal responses) was available and therefore precluded error analysis.

Scoring reliability

Twenty-five percent of the responses from 25% of PWA were scored for interrater and intrarater reliability in distinguishing correct from incorrect responses. Kappa coefficient was 0.98 for intrarater agreement and 0.90 for interrater agreement, indicating high levels of rater reliability.

DATA ANALYSIS AND RESULTS

Table 3 displays descriptive characteristics of the PWA and NC groups. The groups were similar in age, $p = .77$; education level, $p = .07$; and handedness, $p = .20$; however, there was a significant difference in percentage of woman. With that said, the authors are unaware of literature supporting the notion that gender affects oral reading. The aphasia group mean WAB-AQ score was 73.10/100 ($SD = 25.23$) and the aphasia group mean BNT score was 31.62/60 ($SD = 19.70$). The average months poststroke onset was 70.55 ($SD = 54.99$). Based on WAB-AQ classification, there were 56 individuals with anomic aphasia, 16 with Broca's aphasia, 14 with conduction aphasia, 7 with Wernicke's aphasia, 3 with transcortical motor aphasia, and 3 with global aphasia.

Overall reading performance for all participants

As a group, the 99 PWA performed quite variably on the reading stimuli demonstrating a wide range of reading abilities with scores ranging from 0 to 51 (max score = 52) with an average score of 25.56 ($SD = 16.00$). The 29 NC also demonstrated variability, although to a much lesser extent, with scores ranging from 35 to 49 with an average score of 44.97 ($SD = 4.14$).

TABLE 3
Descriptive characteristics for persons with aphasia (PWA) and normal controls (NC)

	N	*Age mean (SD)*	*Education mean (SD)*	*Gender*	*Handedness*
PWA	99	62.02 (13.20)	14.76 (2.65)	63 M, 36 F	91 R, 6 L, 2 A
NC	29	62.79 (10.48)	15.76 (2.47)	9 M, 20 F	29 R
p-value		.77*	.07*	.0026**	.20**

*Independent samples *t*-tests.
**Fisher's exact test. Handedness: right versus nonright.

Frequency of alexia

To determine the frequency of alexia in our convenience sample of 99 PWA, we operationally defined alexia by a score of >2 *SD* below the mean reading score of the NC group (M = 44.97; SD = 4.15). Therefore, within the group of PWA, a score of <36.67/52 on Subtest 1 of the SAPA indicated presence of alexia in this study. We operationalised performance 2 *SD* below the NC mean as a proxy of reading impairment since a NC ceiling effect was not present, and a 2 *SD* criterion has been used in a previous alexia study (Rapcsak et al., 2009). Additionally, a 2 *SD* cut-off is commonly used in test manuals to define impairment (e.g., Comprehensive Aphasia Test; Swinburn, Porter, & Howard, 2004). Of the 99 PWA, 67 individuals (67.78%) scored more than 2 *SD* below the NC mean (see Figure 1). The 32 PWA who did not meet our operationalised criteria for alexia (i.e., scored within 2 *SD* of the NC mean) were not included in the analyses discussed later.

Nature of reading performance for PWA and cooccurring alexia

Reading accuracy for the PWA and alexia group and the NC group is displayed in Figure 2. Overall, the NC group read with an average accuracy of 86.47% (SD = 7.97%) and the PWA and alexia group read with an average accuracy of 33.09% (SD = 23.86%) across all word types combined (regular, irregular, pseudohomophone, and nonword).

To determine whether there was an effect of group and word type, we conducted a 2 (group: alexia versus NC) × 4 (word type: regular, irregular, pseudohomophone, and nonword) mixed model analysis of variance. Results showed significant main effects of group, $F(1, 94) = 162.02$, $MSE = 0.15$, $p < .001$, partial $\eta^2 = 0.63$, significant main effects of word type, $F(3, 282) = 91.74$, $MSE = 0.03$, $p < .001$, partial $\eta^2 = 0.47$, and a group × word type interaction, $F(3, 282) = 2.86$, $MSE = 0.03$, $p = .047$, partial $\eta^2 = .03$. Planned contrasts showed that PWA and alexia read regular words, $t(94) = -16.33$, $p < .001$, irregular words, $t(94) = -11.67$, $p < .001$, pseudohomophones, $t(94) = -13.96$, $p < .001$, and nonwords, $t(94) = -16.20$, $p < .001$, significantly worse than the NCs (Figure 2). Both groups

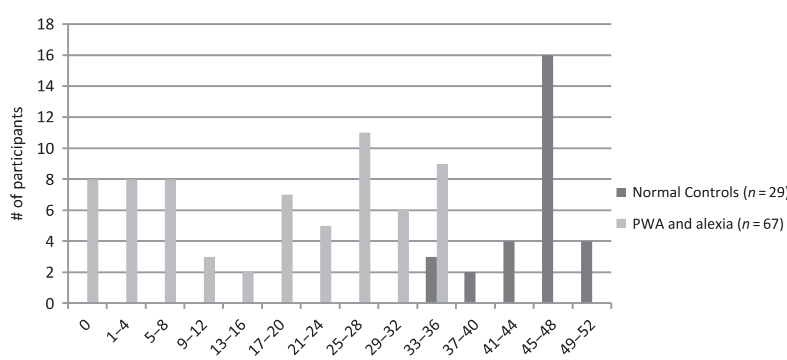

Figure 1. Distribution of reading scores for normal controls and persons with aphasia (PWA) and alexia. SAPA = Standardised Assessment of Phonology in Aphasia (Kendall et al., 2010).

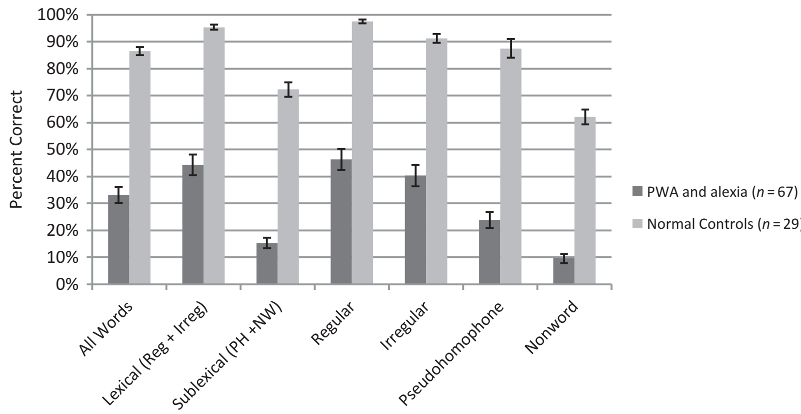

Figure 2. Percent correct oral reading on all word types, lexical words, sublexical words, regular, irregular, pseudohomophone (PH), and nonword (NW) stimuli on the SAPA for normal controls (n = 29) and persons with aphasia (PWA) and alexia (n = 67). SAPA = Standardised Assessment of Phonology in Aphasia (Kendall et al., 2010); error bars = *SE*.

demonstrated the same reading pattern with regular word accuracy > irregular word accuracy > pseudohomophone accuracy > nonword accuracy; however, the PWA and alexia showed this pattern to much a greater extent than the NC group. Despite impairment across all word types, lexical reading abilities (regular and irregular word reading scores) were significantly higher than sublexical reading abilities (pseudohomophones and nonwords reading scores), $t(66) = 9.09$, $p < .001$, for PWA and alexia.

In order to determine the most common subtype of alexia, sublexical (i.e., phonologic and deep) or lexical (i.e., surface), in this sample of PWA and alexia, the ratio of nonword reading/irregular word reading was calculated for each individual with alexia and compared to the NC group mean ratio (.68) (*SD* = .18). If one defines surface alexia by better performance with words read predominately by the sublexical route (nonwords) compared with words read predominately by the lexical route (irregular), and taking into account the performance of our NC sample, only two PWA and coexisting alexia, or 2.99%, had a nonword/irregular word ratio greater than the NC mean ratio and might be characterised as having lexical alexia (see Figure 3). These two PWA had nonword/irregular word ratio scores of 1.22 and 0.92, respectively, indicating better sublexical than lexical reading. The remaining 65 PWA and alexia had nonword/irregular word ratios less than the NC group's ratio and demonstrated superior lexical reading. Moreover, alexia in this sample of PWA appears sublexical in nature, as demonstrated by 66 of the 67 (98.5%) PWA and coexisting alexia being at or below chance level performance for sublexical reading (see Figure 3).

The relationship between aphasia type and alexia type was explored (see Figure 4). In the group of PWA and alexia, there were 25 individuals with anomic aphasia, 16 with Broca's aphasia, 14 with conduction aphasia, 6 with Wernicke's aphasia, 3 with transcortical motor aphasia, and 3 with global aphasia. From the original group of 99 PWA, 31 individuals with anomic aphasia and 1 individual with Wernicke's aphasia did not meet our criteria for alexia. The individual with Wernicke's aphasia scored 37/52, missing the alexia cut-off score (36.68) by one point. Across aphasia

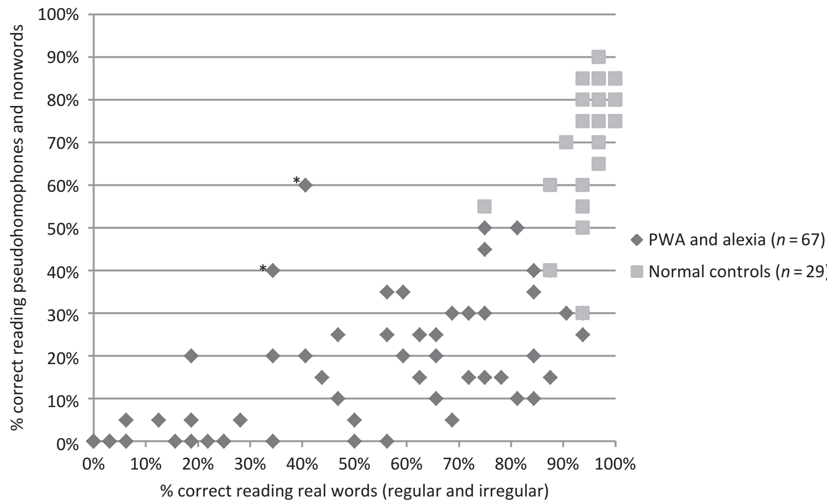

Figure 3. Individual comparison of real word and nonword oral reading abilities for normal controls and persons with aphasia (PWA) and alexia on the SAPA. *PWA and alexia with better sublexical than lexical reading relative to normal controls; SAPA = Standardised Assessment of Phonology in Aphasia (Kendall et al., 2010).

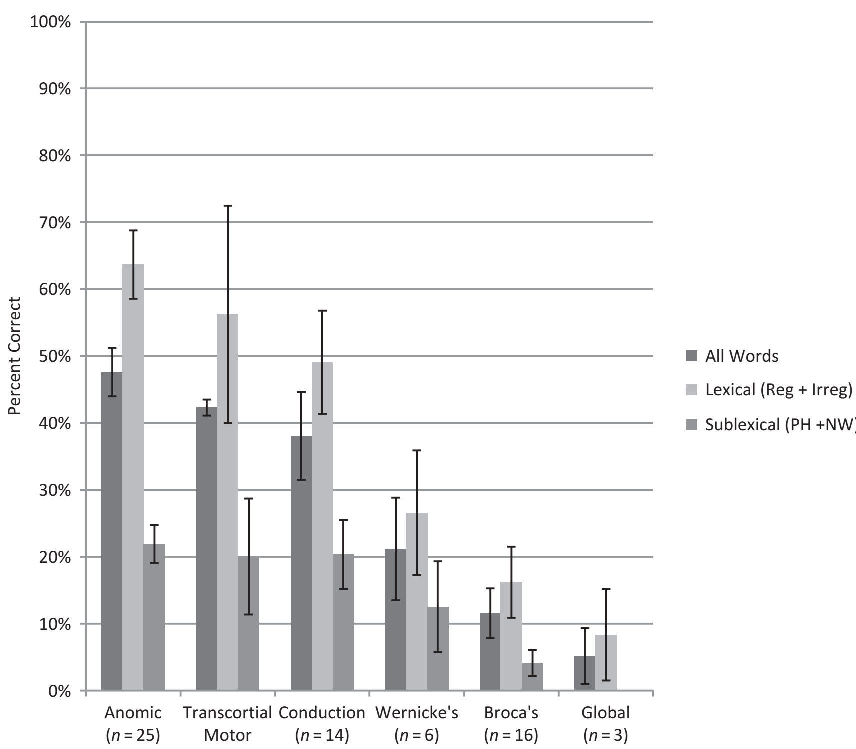

Figure 4. Reading accuracy by aphasia type for 67 individuals with alexia. Aphasia types are based on the WAB-AQ classification system; error bars = *SE*; the individuals with global aphasia earned 0% correct on the sublexical items.

types, there was more variability in lexical (8–64% accuracy) than sublexical (0–22%) reading accuracy, with PWA of all aphasia types performing much worse on sublexical reading items (pseudohomophones and nonwords).

Predictors of oral reading performance

Due to the retrospective nature of this study, we were unable to explore an exhaustive list of potential predictors of alexia; instead, we utilised existing available data to investigate if years of education and aphasia severity might inform reading performance in PWA and alexia. Three simultaneous multiple linear regression analyses were conducted to determine if years of education and aphasia severity (defined by WAB-AQ score) were predictive of reading performance for the 67 PWA with alexia on reading performance for all word types, for performance on real-word reading, and then for performance on pseudohomophones and nonwords. The three analyses were carried out to determine if there was a differential impact of education and aphasia severity on lexical and sublexical reading abilities.

The linear regression models are summarised in Table 4. Overall, the predictors (years of education and aphasia severity) accounted for a significant amount of the variance in all three models: Model 1 (total reading), R^2 = .61, $F(2, 64) = 49.49$, $p < .001$, R^2 adjusted = .60; Model 2 (lexical reading), R^2 = .60, $F(2, 64) = 48.28$, $p < .001$, R^2 adjusted = .59; and Model 3 (sublexical reading), R^2 = .36, $F(2, 64) = 17.87$, $p < .001$, R^2 adjusted = .34.

Performance on WAB-AQ had a unique, positive effect on total reading, lexical reading, and sublexical reading. Holding years of education constant, the models predicted for every one-point increase on WAB-AQ, there is an expected 0.7% increase on total reading score, $p < .001$, $sr^2 = .57$, effect size; an expected 0.9% increase on lexical reading, $p < .001$, $sr^2 = .57$; and an expected 0.3% increase on sublexical reading, $p < .001$, $sr^2 = .30$. Education, on the other hand, was not uniquely predictive of total reading, lexical reading, or sublexical reading, $p = .07$, $p = .14$, $p = .06$, respectively.

TABLE 4
Simultaneous multiple linear regression model results for 67 PWA and alexia

	b	*SE*	*t*	*p-Value*	*β*
SAPA total reading					
Education	0.012	0.007	1.84	.070	0.15
WAB-AQ	0.007	0.001	9.61	<.001*	0.76
SAPA regular + irregular words					
Education	0.013	0.009	1.51	.137	0.12
WAB-AQ	0.009	0.001	9.57	<.001*	0.76
SAPA pseudohomophones + nonwords					
Education	0.011	0.006	1.88	.064	0.19
WAB-AQ	0.003	0.001	5.51	<.001*	0.56

SAPA = Standardised Assessment of Phonology in Aphasia (Kendall et al., 2010). WAB-AQ = Western Aphasia Battery Aphasia Quotient (Kertesz, 1982).

DISCUSSION

The purpose of this study was to provide initial evidence regarding the frequency and nature of alexia in a convenience sample of 99 persons with aphasia (PWA) and, additionally, to determine if level of education and severity of aphasia were predictive of oral reading abilities in PWA and coexisting alexia. Overall, we found that the majority (68%) of PWA in this sample met our criteria for alexia (i.e., performing <2 *SD* below the NC group). This finding is consistent with previous work that has recognised the common occurrence of reading impairment in PWA (Beeson & Hillis, 2001; Cherney, 2004; Riley & Kendall, 2013; Webb & Love, 1983). Furthermore, we found that the alexia type tended to be sublexical in nature, as evidenced by poorer sublexical reading (pseudohomophones and nonwords) compared to lexical reading (regular and irregular words) with 66 out of 67 (98.5%) of the PWA with alexia being at or below chance level performance for sublexical reading (see Figure 3). In addition, we found a wide range of aphasia types present in this group of individuals of alexia (see Figure 4), with more variability in lexical (8–64% accuracy) than sublexical (0–22%) reading accuracy across aphasia types. Moreover, aphasia severity was found to be significantly predictive of oral reading performance with higher WAB-AQ scores predicting less severe alexia for real words and nonwords.

Our finding that 68% of PWA demonstrated evidence of alexia is in contrast with previous work that found reading impairment always occurs with aphasia (Duffy & Ulrich, 1976; Webb & Love, 1983). This discrepancy may be attributed to differences in the stimuli used to assess reading abilities and the operational definition of alexia employed in this study. In contrast to the four word types (i.e., regular, irregular, pseudohomophone, and nonword) assessed in this study, these earlier studies examined reading through use of visual recognition, comprehension, and real word and sentence oral reading tasks (Webb & Love, 1983) or via clinical interviews and standardised aphasia tests (Duffy & Ulrich, 1976). It is also possible that our operational definition of alexia is more conservative than criteria employed in these prior works. In other words, PWA in the present study could demonstrate reading difficulties and not be identified with alexia if they scored within 2 *SD* of the NC group mean.

The HF of alexia in PWA reported in this study is consistent with connectionist language models (Nadeau, 2001; Plaut, 1999; Plaut et al., 1996) that propose neuronal language representations needed for successful oral reading do not exist in isolation, but rather are distributed across the language dominant hemisphere. Therefore, there is a high probability that a stroke damaging the neuronal substrate for spoken language will also damage, to some degree, the substrate for reading aloud, with the extent of concurrent impairment depending to some degree on lesion locus and extent. These structurally distributed language networks are thought to degrade gracefully when damaged. Our finding that the PWA and alexia read in a similar (yet impaired) pattern to the NC (i.e., regular words > irregular words > pseudohomophones > nonwords) is suggestive of a graceful degradation of reading function. Graceful degradation reflects the fact that knowledge is encoded in synaptic connections through a spatially distributed neural network. Thus, damage to some portion of that network increases the probability of error or nonresponse, but it does not fundamentally transform the nature of the remaining knowledge encoded in the network.

PWA and alexia demonstrated significantly worse reading than the NC on all word types and superior reading of lexical items (regular and irregular words) relative to sublexical items (pseudohomophones/nonwords). Only 2 of 67 (2.99%) PWA and alexia were classified as having a lexical/surface alexia (defined by a ratio of nonword/irregular word reading greater than the mean ratio for the NC), suggesting that most of the PWA and alexia in this study present with a sublexical alexia. However, it is critical to note that the somewhat variable reading performance of the NC group (see Figure 3) and lack of access to the participant's verbal reading responses preclude a sharp distinction between alexia subtypes in this sample.

We offer three possible explanations for this tentative evidence of a tendency towards sublexical alexia in PWA and alexia. First, it is possible that some PWA and alexia may have possessed premorbid developmentally weaker phonological processing abilities, and it is likely these individuals were more susceptible to phonologic impairment after left hemisphere stroke. Second, we know that PWA in general, regardless of how well developed their phonological representations may have been prior to brain damage, often demonstrate impairment in phonological awareness and processing abilities reflecting loss of phonologic sequence knowledge (Blumstein, Baker, & Goodglass, 1977; Kendall et al., 2008; Nadeau, 2001). Loss of phonological sequence knowledge, combined with interruption of connections between the substrates for phonology and orthography, renders grapheme sequence to phoneme sequence conversion difficult or impossible, depending on extent of brain damage. Third, some PWA and alexia may show impaired sublexical reading abilities due to a reliance on language representations in the right hemisphere during reading tasks to compensate for damaged language representations in the left hemisphere.

The right hemisphere hypothesis (Coltheart, 2000) postulates that the right hemisphere is capable of reading, albeit with a bias towards reading of highly imageable real words (Coslett, 2000). Therefore, the right hemisphere hypothesis accounts for our finding of more impaired sublexical reading abilities in PWA as the right hemisphere is thought to substantially lack the substrate for phonological sequence knowledge needed to read nonwords and is better equipped to support whole word reading via the lexical route (Zaidel, Iacobini, Berman, Zaidel, & Bogen, 2011).

Regression analyses showed that together aphasia severity (measured by WAB-AQ) and years of education accounted for a significant amount of the variance in lexical and sublexical reading combined (60%), lexical reading only (60%), and sublexical reading only (36%). However, only aphasia severity was shown to be a significant, unique predictor of oral reading abilities in this sample of PWA and coexisting alexia. Specifically, reading accuracy for real words and nonwords was predicted to increase as aphasia severity decreased (as indicated by improved score on the WAB-AQ). It is noteworthy that the regression models accounted for a greater amount of the variance in real-word reading than nonword and pseudohomophone reading. This finding is likely attributable to the fact that *both* semantic and phonologic knowledge contribute to reading real words, whereas the reading of nonwords relies on the integrity of phonological sequence knowledge and connections to its substrate from the substrate for orthography. Our examination of aphasia subtypes present in this sample of individuals with alexia provides some support for these regression predictions. Figure 4 illustrates that individuals with aphasia types associated with higher WAB-AQ scores (e.g., anomic aphasia and conduction aphasia) read more accurately than those individuals with aphasia types associated with lower WAB-AQ scores (e.g., Broca's aphasia and global aphasia).

We found only a trend relationship between years of education and alexia severity. Other studies have found a significant relationship, suggesting that higher education may act as a protective cognitive reserve in individuals with aphasia (Morelli et al., 2004; Smith, 1971; Webb & Love, 1983) and individuals with dementia (Stern, Alexander, Prohovnik, & Mayeux, 1992; Stern et al., 1994). We propose three explanations for our contrasting finding. First, our study may have been underpowered to detect such an effect. Second, it is possible that our group of PWA and alexia did not have enough variability in their years of education for this variable to significantly predict reading. All but two (97%) of the individuals with alexia in this study had at least a high school education with many having achieved college degrees and a few achieving graduate degrees. Webb and Love (1983) analysed reading abilities from a more educationally heterogeneous group of PWA and found a significant difference in reading between those individuals with and without a high school education, with superior reading by those with a high school degree. Third, we might have achieved different regression results had we used different stimuli. Our regression findings can only speak to the relationship between years of education and reading performance on the 52 reading items on the SAPA (see Table 1).

Clinical implications

Our findings have clinical relevance regarding aphasia-associated alexia. The results indicate that alexia is frequent among individuals with chronic aphasia (of various subtypes) and therefore may lend support for routine assessment of reading abilities in this population. Specifically, our findings suggest that assessing sublexical reading abilities, in addition to the more common assessment of lexical reading abilities, may be beneficial. Assessing both sublexical and lexical reading abilities via oral reading of regular, irregular, pseudohomophone, and nonwords may potentially reveal information regarding alexia subtype and underlying semantic, phonologic, and orthographic capabilities that may inform rehabilitation.

Limitations and future directions

Due to the retrospective design of this study, there are limitations regarding participant and stimuli selection that undoubtedly influenced our findings and deserve discussion. We employed a retrospective analysis of a convenience sample of individuals with chronic aphasia that may not have been fully representative of the population with stroke-associated aphasia at large, despite our inclusion of individuals with a range of aphasia types and severity, and despite the fact that type of aphasia and alexia was not a criterion for inclusion. The inclusion of a wide range of aphasia types and severity was potentially problematic given that some individuals in this sample were likely to present with cooccurring motor speech impairments, which could have negatively impacted oral reading performance.

We are unable to account for the exact number of individuals in our study that presented with apraxia of speech (AOS) or dysarthria since results from motor speech evaluations were not available. However, data from individuals with known severe AOS or dysarthria were not included in the analyses. Moreover, many of the speech errors (e.g., distortions, pauses, and prolonged rate) commonly made by individuals with AOS would have been scored as correct since only apparent omissions, semantic errors, or phonologic errors (additions, substitutions, etc.) were scored as incorrect.

With that said, we cannot ignore the fact that it is often difficult to determine the difference between an apraxic error and a phonologic error. The mis-scoring of phonemic slips (phonologic error) as phonemic distortions (apraxic error), to the extent that they occurred, would have led to underestimation of the severity of reading impairment (sensitivity issue). On the other hand, mis-scoring of phonemic distortions as phonemic slips would have led to an overestimation of severity of reading impairment (specificity issue). In future alexia research, an effort should be made to objectively identify those individuals with AOS and either exclude them from the alexia study or better yet include their data and investigate reading impairment in individuals with apraxia and aphasia compared to those individuals without motor speech difficulties.

Regarding the stimuli employed, this study is limited by the fact that only oral reading abilities for single words were assessed, and this work does not speak to text comprehension abilities in PWA and alexia. Our analysis was based solely upon performance on a subtest of an instrument (i.e., SAPA) probing particular reading pathways (lexical and sublexical). The number and types of reading items on the SAPA are less exhaustive than would be expected for stimuli designed a priori to assess alexia. With that said, we believe reading performance on these stimuli can still provide valuable insight into an individual's reading capabilities. In future work, expanding the stimuli and including error analyses would allow for more specific alexia classifications to be made.

In summary, these retrospectively obtained results suggest that the majority of individuals with chronic aphasia in this convenience sample present with acquired reading difficulty. Based on the small number of stimuli employed, the severity of the reading difficulty in PWA and alexia appears varied (see Figure 3). However, there appears to be consistent differential difficulty for words requiring sublexical processing compared to lexical processing, regardless of aphasia type or severity. This finding is supported by more severe impairment of obligatorily sublexical reading (i.e., pseudohomophones/nonwords) abilities compared to lexical reading (i.e., real words) abilities for most of the PWA and coexisting alexia in this sample. Future a priori work is needed to provide greater control over participant and stimuli selection to more systematically explore the presence, nature, and predictors of alexia in chronic aphasia.

Manuscript received 29 November 2013
Manuscript accepted 2 July 2014
First published online 6 August 2014

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