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# Research Article

# Treatment for Alexia With Agraphia Following Left Ventral Occipito-Temporal Damage: Strengthening Orthographic Representations Common to Reading and Spelling

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Purpose: Damage to left ventral occipito-temporal cortex can give rise to written language impairment characterized by pure alexia/letter-by-letter (LBL) reading, as well as surface alexia and agraphia. The purpose of this study was to examine the therapeutic effects of a combined treatment approach to address concurrent LBL reading with surface alexia/agraphia.

Method: Simultaneous treatment to address slow reading and errorful spelling was administered to 3 individuals with reading and spelling impairments after left ventral occipitotemporal damage due to posterior cerebral artery stroke. Single-word reading/spelling accuracy, reading latencies,

Equired impairments of written language process-<br>ing are relatively common after damage to left<br>ventral occipito-temporal (lvOT) cortex and un-<br>derlying white matter. The associated syndromes include ing are relatively common after damage to left ventral occipito-temporal (lvOT) cortex and underlying white matter. The associated syndromes include letter-by-letter (LBL) reading, surface alexia, and surface agraphia (Déjerine, 1892; Epelbaum et al., 2008; Rapcsak & Beeson, 2004; Sebastian et al., 2014). LBL reading is the typical compensatory strategy observed when there is disconnection or disruption of visual input to critical regions for orthographic processing (Binder & Mohr, 1992; Damasio & Damasio, 1983). LBL reading is often characterized as pure alexia, or alexia without agraphia, when

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and text reading were monitored as outcome measures for the combined effects of multiple oral re-reading treatment and interactive spelling treatment. Results: After treatment, participants demonstrated faster and more accurate single-word reading and improved text-

reading rates. Spelling accuracy also improved, particularly for untrained irregular words, demonstrating generalization of the trained interactive spelling strategy. Conclusion: This case series characterizes concomitant LBL with surface alexia/agraphia and demonstrates a successful treatment approach to address both the reading

and spelling impairment.

spelling knowledge is preserved. In some cases, however, the serial LBL reading profile is accompanied by an impairment of orthographic knowledge, so that reading and spelling are accomplished with overreliance on grapheme– phoneme and phoneme–grapheme conversion, respectively (Bowers, Arguin, & Bub, 1996; Friedman & Hadley, 1992; Patterson & Kay, 1982). In other words, LBL reading may co-occur with surface alexia and surface agraphia as depicted in Figure 1a. For reading, the identification of the

graphemic sequence comprising a word does not necessarily lead to accurate word recognition, and further decoding is achieved by letter-to-sound conversion. This sublexical reading approach is prone to error for irregularly spelled words, such as blood, which may ultimately be pronounced to rhyme with rude (see Figure 1a). Degraded orthographic representations also affect spelling, so that phonologically plausible errors are common for irregularly spelled words, as in spelling blood as blud.

From a neural perspective, LBL reading has been associated with damage to white matter pathways linking visual processing areas to cortical regions that store orthographic memory representations of familiar words (Damasio

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Figure 1. (A) A cognitive model of written language processing depicting disruption of visual input to the orthographic lexicon (hash marks), an alternate letter-by-letter reading route (dashed lines), and graying of the orthographic lexicon to indicate damage. (B) Schematic depiction of the presumed effect of behavioral treatments, showing multiple oral re-reading treatment (MOR Tx) promoting whole word processing rather than letter-by-letter reading, and interactive spelling treatment promoting combined reliance on lexical and sublexical processing of information to support word recognition and spelling.



& Damasio, 1983; Déjerine, 1892; Geschwind, 1965), or with damage to a left midfusiform region referred to as the visual word form area (VWFA; Binder & Mohr, 1992; Cohen et al., 2003; Leff et al., 2001; Pflugshaupt et al., 2009; Roberts et al., 2013; Yeatman, Rauschecker, & Wandell, 2013). The critical role of the VWFA in central orthographic processing is supported by demonstrations that this region shows overlapping patterns of activation in functional imaging studies of reading and spelling in normal subjects (Beeson et al., 2003; Bruno, Zumberge, Manis, Lu, & Goldman, 2008; Kronbichler et al., 2007; Rapp &

Lipka, 2011; also see Price, 2012 for a recent review). These observations suggest that the VWFA is the likely neural substrate of the orthographic lexicon postulated by cognitive models of written language processing (Hillis, 2002; Purcell, Turkeltaub, Eden, & Rapp, 2011; Rapcsak & Beeson, 2004; Rapp, 2002; Sebastian et al., 2014). Consistent with this notion, damage to this region has been associated with a loss of orthographic knowledge as observed in surface alexia and surface agraphia (Friedman & Hadley, 1992; Rapcsak & Beeson, 2004; Samuelsson, 2000; Tsapkini & Rapp, 2010).

The co-occurrence of LBL reading with a surface alexia pattern was described in individual cases by Patterson and Kay (1982) as Type 2 LBL and by Friedman and Hadley (1992) as LBL surface alexia. Although surface agraphia has also been evident in such individuals (e.g., Behrmann & McLeod, 1995; Friedman & Hadley, 1992; Patterson & Kay, 1982; Rapp & Caramazza, 1991), the concomitant spelling impairment has received limited attention. This is surprising because spelling impairment tends to be more severe and persistent than reading in cases of surface alexia/agraphia, due to greater task difficulty (Rapcsak & Beeson, 2004; Sebastian et al., 2014). There are treatment protocols that demonstrate positive outcomes directed toward LBL reading and surface alexia/agraphia independently, but to our knowledge, no treatment studies have addressed these concomitant reading and spelling difficulties.

Multiple oral re-reading (MOR) is a text-based treatment approach that has been used to remediate LBL reading (Beeson, 1998; Beeson, Magloire, & Robey, 2005; Lacey, Lott, Snider, & Sperling, 2010; Moody, 1988; Moyer, 1979; Tuomainen & Laine, 1991). MOR involves repeated reading aloud of the same text until a criterion (usually a target reading rate) is reached. As depicted in Figure 1b, the treatment is intended to shift the individual away from the LBL reading strategy and to strengthen links between visual input and the associated orthographic representations.<sup>1</sup> Repeated practice reading words within a semantic/syntactic context provides top–down support to reduce the effects of the peripheral processing impairment (Beeson et al., 2005; Moyer, 1979; Tuomainen & Laine, 1991). However, the ultimate goal of MOR treatment is to increase bottom-up processing so that word recognition improves, and to promote interactive processing of all available information to support reading. The supported reading stimulation from MOR has a rehabilitative effect so that reading rate and accuracy are better for untrained text, and word-form recognition improves as evidenced by a reduced word-length effect. A recent review of MOR treatment by Starrfelt, Olafsdóttir, and Arendt (2013) examined treatment outcomes from seven cases reported in four studies since 1990 (Beeson, 1998; Beeson et al., 2005; Lacey et al., 2010; Tuomainen & Laine, 1991). Four of the cases showed significant improvement in reading speed for words and/or text, and one individual (RB in Beeson et al., 2005) showed a significant improvement in reading rate for unfamiliar text and a resolution of the word-length effect by the end of treatment. These findings are encouraging, but highlight the need for more research regarding this approach.

Treatments for central orthographic impairments include retraining specific lexical items, particularly irregularly spelled words, which are prone to error due to overreliance on phonological strategies for reading and spelling. For surface alexia, irregular words have been trained using mnemonic semantic cues (Byng & Coltheart, 1986), paired homophones (Scott & Byng, 1989), or repeated whole-word reading (Friedman & Robinson, 1991). Treatments for surface agraphia have included several approaches that support the relearning of spelling of irregularly spelled words with reliance on visual imagery (de Partz, Seron, & van der Linden, 1992), mnemonics (Schmalzl & Nickels, 2006), paired homophones (Behrmann, 1987), and repeated study and recall of targeted words (Rapp, 2005). Such itemspecific approaches have good outcomes for relearning trained items, but limited generalization to untrained words. In contrast, an interactive approach to spelling treatment has been used to train a problem-solving strategy that promotes use of sublexical (i.e., phoneme–grapheme conversion) and residual lexical spelling abilities (Beeson, Rewega, Vail, & Rapcsak, 2000; Beeson, Rising, & Rapcsak, 2011). In this approach, individuals are instructed to generate phonologically plausible spellings for words they cannot spell. Participants then evaluate spellings and practice detecting and correcting errors, using an electronic spell-checker to provide corrective feedback as needed. The focus of treatment is depicted in Figure 1b as strengthening lexical and sublexical processing and the interaction of all available linguistic information (orthographic, phonological, and semantic) for reading and spelling. Evidence for interactive processing of lexical and sublexical information was demonstrated by error responses of an individual with dysgraphia following brain injury reported by Rapp, Epstein, and Tainturier (2002). This individual made spelling errors such as KNOLIGE for knowledge, which includes the low-frequency kn for /n/ derived from lexical knowledge combined with a phonologically plausible ending lige. Beeson, Rising, Kim, and Rapcsak (2010) also observed such responses in the context of interactive spelling treatment (e.g., anteeque for antique).

Given that MOR and interactive spelling treatments have been used independently to address reading and spelling impairments in individuals with acquired alexia and agraphia, it is reasonable to expect that a combined approach may result in written language improvements in both domains. In this study, we present three individuals with acquired alexia and agraphia associated with lvOT damage due to stroke. They were similar to one another in that each had significant word length effects in reading co-occurring with surface agraphia. In order to address these impairments, we implemented a concurrent treatment approach intended to improve the speed of visual word recognition (using MOR) and to strengthen orthographic representations (using interactive spelling treatment). We expected that this combined approach would improve both peripheral and central written language impairments observed in these individuals.

## Method

Three individuals with acquired language impairment following stroke participated in this study, and their performance was compared with that of 10 healthy adults

<sup>&</sup>lt;sup>1</sup>MOR and other oral reading treatments have also been implemented in individuals with reading deficits associated with aphasia and phonological impairment due to left perisylvian damage (e.g., Beeson & Insalaco, 1998; Cherney, Merbitz, & Grip, 1986; Lacey et al., 2010), but that is not the focus of this article.

on various tasks. The research protocol was approved by the Human Subjects Protection Program at the University of Arizona, and all participants provided informed consent.

### **Participants**

#### Participant 1

Participant 1 (P1) was an 80-year old, right-handed woman with 12 years of education who experienced a left posterior cerebral artery (PCA) stroke and was evaluated at 2 weeks, 4 months, and 8 months poststroke. She was a monolingual English speaker who worked as a receptionist prior to her retirement. P1 reported that prior to her stroke she had excellent reading and spelling skills (each rated 5 on a 5-point scale).

#### Participant 2

Participant 2 (P2) was a 73-year old, right-handed man with 14 years of education who was 9 years poststroke. He was an English/Spanish bilingual speaker who used English as his primary language. He had a history of two strokes: a small right occipital stroke (right PCA) and a left PCA stroke that resulted in his written language difficulties. P2 was able to read printed stimuli presented within his preserved field of view, using his finger to mark his place while reading. His restricted field of view was a poor prognostic sign for improving reading speed, but P2 was highly motivated to maximize recovery of his written language skills, which he judged to be excellent prior to his stroke (5 on 5-point scale for both reading and spelling).

#### Participant 3

Participant 3 (P3) was a 76-year old, right-handed man with 14 years of education who was 10 months poststroke. He was a monolingual English speaker, and he was retired from his job as a machinist. Medical history indicated that he had suffered two strokes within a few months of each other. The first resulted in damage to left posterior middle frontal gyrus (Brodmann Area 6), with no reported language difficulties or behavioral changes. The second stroke in left PCA distribution resulted in marked written language impairment. Similar to P1 and P2, P3 indicated good reading and spelling skills (5 out of 5 rating for both) prior to his stroke.

#### Control Participants

A group of 10 individuals (two men, eight women; matched for age and education to the participants with alexia with agraphia) with no history of neurological conditions or developmental language impairments provided normative data for some study tasks. The performance of the individuals with alexia/agraphia was compared with controls using modified  $t$  tests for comparing single cases against small groups (Crawford & Howell, 1998), except in cases where there was no variance in control group performance (e.g., ceiling effects).

#### Lesion Analysis

Structural brain scans were obtained from clinical records for all participants, and additional research scans were obtained for P1. All available images (computed tomography and/or magnetic resonance imaging scans using T1, T2, FLAIR, or diffusion weighted imaging) were used to optimize the ability to visualize and accurately map the lesions of each participant. Using the procedures detailed in Andersen, Rapcsak, and Beeson (2010), regions of pathology were digitally drawn by hand on each participant's brain scan in native space and then the scans and lesion maps were warped to a standard template brain for display. As shown in Figure 2, P1 and P2 had relatively similar lesion location and extent affecting left medial occipital cortex and fusiform gyrus, including damage to the VWFA. Damage to primary visual cortex was less extensive for P1, who had a right upper quadrantanopia. P2 had a right homonymous hemianopia, as well as a left lower quadrantanopia due to a right medial supracalcarine lesion located above the level of the axial slices displayed in Figure 2. P3 had damage to left medial temporal lobe regions that extended posteriorly into midfusiform cortex encroaching on the VWFA. The lesion did not affect primary visual cortex, and thus he did not have a visual field defect.

#### Behavioral Assessment

All three participants were administered the Western Aphasia Battery (Kertesz, 1982) and the Boston Naming Test (BNT; Kaplan, Goodglass, & Weintraub, 2001) to examine spoken language comprehension and production. The picture version of Pyramids and Palm Trees (Howard & Patterson, 1992) was administered to assess nonverbal semantic knowledge. Subtests from the Psycholinguistic Assessment of Language Processing in Aphasia (18, 23, 26; Kay, Lesser, & Coltheart, 1992) were administered to assess letter recognition and to confirm sufficient vision for perception of written stimuli. Phonology–orthography relations were assessed using sound–letter/letter–sound transcoding tasks.

The Arizona Battery for Reading and Spelling (ABRS; Beeson & Rising, 2010), a list of 40 regular and 40 irregular words (balanced for frequency, length, and imageability) and 20 pronounceable nonwords, was administered to examine lexical and sublexical reading and spelling abilities. Responses were scored for accuracy, and the magnitude of the regularity effect (regular–irregular) was calculated for reading and spelling.

The influence of word-length on reading was assessed using the Arizona Word Length List (Beeson, n.d.), a 160-item list containing 40 words at four, five, six, and seven letters. Words on this list were balanced for frequency, imageability, regularity, and grammatical class, and were presented via Direct RT (Jarvis, 2001) on a laptop computer. Response times (RTs) in milliseconds were recorded from stimulus onset to initiation of voice response, and RT analyses were computed for correct responses only, with outliers

Figure 2. Lesion maps for the three participants (left); average reading reaction times for four- to seven-letter words, with standard deviation bars (middle); reading and spelling performance on regular and irregular words (right). Lesions are normalized to standard MNI template depicting damage to left ventral occipito-temporal (lvOT) regions at z = −17 (leftmost) and z = −12. Red circles are 12 mm spheres indicating the location of the visual word form area (VWFA) derived from a meta-analysis of functional imaging studies of reading (Jobard, Crivello, & Tzourio-Mazoyer, 2003), and represent  $x = -44$ ,  $y = -58$ ,  $z = -15$ . \*Significant regularity effect ( $p < .05$ ).



(+/− 2 SDs of mean) removed. The word length effect, represented by the slope of RT increase from four to seven letters, was calculated and reported in milliseconds per letter; correlations were calculated between RT and word length.

Text reading was assessed using the Gray Oral Reading Test–3 (GORT–3; Wiederholt & Bryant, 1992) and two additional 100-word passages at 1.6 and 12.8 grade levels according to Flesch–Kincaid calculations (Kincaid, Fishburne, Rogers, & Chissom, 1975). Measures of reading rate (words per minute [wpm]) and accuracy (correct words minus errors per 100 words) were computed for each passage. Alternate forms of the GORT–3 were administered preand posttreatment, with each participant reading paired passages of equivalent difficulty level before and after treatment.

## Pretreatment Profiles

#### **P1**

P1 was initially evaluated at 2 weeks poststroke, but additional health concerns prevented her from beginning treatment until 8 months after her stroke. This delay allowed the opportunity to document aspects of natural language recovery at 2 weeks, 4 months, and 8 months poststroke. At all three time points, P1's spoken language profile was consistent with anomic aphasia on the Western Aphasia Battery (Aphasia Quotient  $[AQ] = 89.7$  at 2 weeks poststroke and 90.4 at 4 months poststroke) with considerable naming impairment as measured by the BNT (13/60 at 2 weeks and 20/60 at both 4 and 8 months poststroke). Semantic knowledge was spared and letter perception was within normal limits, as was sound–letter/letter–sound transcoding (Table 1).

At 2 weeks poststroke, P1 demonstrated prominent surface alexia and agraphia. Her oral reading of single words was 76.5% correct overall, characterized by production of phonologically plausible errors for irregular words (e.g., pronouncing pint to rhyme with hint) and a marked regularity effect, 27.5%,  $\chi^2$  (1) = 6.9, p = .009. Spelling accuracy was more impaired than reading (71.5%) and was characterized by phonologically plausible spelling errors (e.g., spelling *bowl* as *BOAL*). When re-evaluated at 4 and 8 months poststroke, overall accuracy of single-word reading had improved (96.5% and 95%, respectively), and the regularity effect in reading had resolved,  $\chi^2$  (1) = 1.39, p = .24 at 4 months poststroke. Single-word spelling accuracy also showed some improvement at 4 months (83%) and 8 months (87.5%) compared with P1's initial performance, but the regularity effect in spelling remained significant, 25% at 8 months,  $\chi^2$  (1) = 9.26, p = .002. Persistent reliance on a sublexical spelling strategy was evident by the preponderance of phonologically plausible errors (e.g., glacier spelled as GLASHUR; prove spelled as PRUVE), as is typical of surface agraphia. Although P1 performed well on phonology– orthography transcoding tasks and nonword spelling, she had some difficulty reading nonwords (80% correct) in relation to real word reading (95% correct; see Table 1).

At each evaluation, P1's reading reaction times were slow relative to controls, with a significant word length

effect. At 8 months, immediately prior to treatment, her average reaction time was 1,365 milliseconds per word with an increase of 141.3 milliseconds per letter (Table 1). Although she was not an overt LBL reader, it was clear that P1 used a serial strategy to decode words to some extent; the correlation between reading reaction time and word length was significant,  $r = .936$ ,  $p = .032$ . As expected, her text reading was slow (around 85 wpm), but was relatively accurate.

#### P2

P2 had anomic aphasia with an aphasia quotient of 89.4, and a score of 42/60 on the BNT. His semantic knowledge was spared. Despite his visual impairment, P2 was able to identify and name all letters of the alphabet without error, and was able to accurately recognize when letters were presented as mirror reversals (Table 1).

P2's single-word reading accuracy was better for regular words compared with irregular words (93% vs. 80%), but this difference did not reach significance,  $\chi^2$  (1) = 1.69,  $p = 0.19$ . When reading aloud, he made phonologically plausible errors (e.g., producing the /b/ in *debt*) and also produced visually similar words (e.g., broad for board). P2's reading reaction time was slow and there was a significant word length effect (see Table 1). Although there was not a prototypical stair-step increase of RT for each letter, and the correlation between word length and RT did not reach significance  $(r = 789, p = .105)$ , P2 read five-, six-, and seven-letter words at a slower rate than four-letter words. He did not demonstrate overt LBL reading. Text reading was slow at about 56 wpm, with about three errors per 100 words. P2's spelling profile was consistent with surface agraphia, with a pronounced regularity effect,  $28\%$ ,  $\gamma^2$  (1) = 5.27,  $p = .02$ . He often relied on a phonological strategy to spell irregular words (e.g., circuit spelled CURCIT, yacht spelled *YAT*), but he also made some errors in letter selection that affected spelling of regular as well as irregular words (e.g., fact spelled FAIT, hunch spelled HANCH). Spelling accuracy for words and nonwords was similar, and P2 performed in the low normal range on the phonology– orthography transcoding tasks.

#### P3

When evaluated at 10 months poststroke, P3 had a very mild anomic aphasia  $(AQ = 91.4)$  and a BNT score of 51/60. His semantic skills were well preserved. P3's singleword reading was fairly accurate overall (91.5% correct), but he was an overt LBL reader with a significant wordlength effect (Table 1). Although P3 was able to name individually presented letters of the alphabet without error, he made letter naming errors when trying to decode written words, leading to errors such as bribe read as "t-r-i-b-e … tribe." P3's reading reaction time was slow (average 2,933 milliseconds per word), as was his text reading rate (approximately 53 wpm). There was a significant wordlength effect for words from four to seven letters,  $r = .951$ ,  $p = .024$ , with a slope of 216 milliseconds per letter. P3 had no regularity effect for reading,  $\chi^2$  (1) = 0.16, p = .69, but had a marked regularity effect for spelling,  $33\%$ ;  $\chi^2$  (1) = 9.3,





Note. P1 = participant 1; P2 = participant 2; P3 = participant 3; WAB = Western Aphasia Battery; PALPA = Psycholinguistic Assessment of Language Processing in Aphasia; RT = response time; wpm = words per minute.

<sup>a</sup>Data from nine control participants. <sup>b</sup>Control participants read the two passages; participants with alexia/agraphia read the same two passages along with the Gray Oral Reading Test–3 passages.

Significantly impaired relative to control data using Crawford and Howell's (1998) modified t test:  $\tau_p < .05; \tau_p < .01; \tau_p < .001$ .

 $p = .002$ . The majority of his spelling errors were phonologically plausible (e.g., choir spelled QUIAR, ghost spelled GOAST). Like the other two participants, P3 had some difficulty reading nonwords (75% correct). Nonword spelling was in the low normal range at 85% correct, and his phonology–orthography transcoding skills were clearly sufficient to support a sublexical strategy for spelling (95% correct overall).

## Summary and Treatment Plan

All three participants with lvOT damage demonstrated a clinical profile of alexia with agraphia that seemed to reflect a combination of peripheral visual and central orthographic processing deficits. The written language impairment was characterized by relatively accurate but slow single-word reading with significant word length effects and surface agraphia. P1 and P2 also showed mild regularity

effects for reading, consistent with surface alexia. All participants shared a strong motivation to improve their written language skills, and they each received individual treatment during hour-long sessions conducted twice per week.

## Reading Treatment

Reading was treated at the text level using the MOR approach as described by Beeson et al. (2005). This treatment targets reading fluency (rate, accuracy) by focusing on repeated re-reading of text (see protocol in Appendix).

#### Procedure

During the reading portion of each treatment session, participants first read a novel text to probe reading rate (wpm) and accuracy (deviations from print per 100 words). Next, a passage was targeted for practice and the initial reading rate and accuracy were measured. Repeated reading

aloud of the practice passage followed, with the clinician maintaining accountability for accurate reading and correction of errors. Daily oral reading homework of the practice passage was assigned for a minimum of 30 min per day, with instructions to complete a log recording the time spent in practice each day. A reading rate of 100 wpm was set as the target rate, but to maintain interest and compliance with treatment procedures, a new passage was assigned after four treatment sessions (2 weeks) even if the criterion was not met.

#### Probe/Practice Stimuli

Probe stimuli were passages of 100 to 150 words, excerpted from the Scientific Research Associates (SRA; 1978) series. Difficulty level of the materials was chosen to approximate 60 to 70 wpm on the basis of initial testing on the GORT–3. Passages of approximately 250 to 300 words (from the SRA series or self-selected texts) were used for re-reading practice. On average, passages for home practice were slightly more advanced than the probe texts by 0.5 to 1 grade level. Reading grade levels for self-selected texts were determined using the Flesch–Kincaid readability statistic calculated in Microsoft Word tools.

#### Outcome Measures

The *direct* measure of treatment was reading rate on trained passages (in wpm), measured at each treatment session. For each passage, the initial reading rate was compared with the last reading rate using paired sample  $t$  tests. Effect sizes for trained passages were calculated from the difference between the average reading rates for the first and last readings across all trained passages divided by the standard deviation of reading rates at first reading (after methods described by Orjada & Beeson, 2005).

To test for generalization, reading rate and accuracy were probed on a novel (i.e., new) passage at the beginning of each treatment session. Accuracy was calculated for the 100-word passages by subtracting the number of errors (deviations from print) from 100 and converting to a proportion (100 – # errors)/100. Probe data from the first three pretreatment and last three treatment sessions were used to calculate effect sizes for rate and accuracy using Busk and Serlin's (1992) *d* statistic (pre- and posttreatment difference divided by the standard deviation of pretreatment probes). Additional measures of generalization included text reading using the alternate version of the GORT–3 and the two 100-word passages read during pretreatment assessment. Pre- and posttreatment values for reading rate and accuracy were tested using paired sample  $t$  tests. To evaluate whether treatment affected word length effect, reading reaction times were sampled using the Arizona Word Length list, and correlations were calculated between word length and reaction times after treatment.

#### Spelling Treatment

#### Procedure

Interactive spelling treatment was administered concurrently with reading treatment during the second half of each session. Participants were trained to implement a three-step problem-solving strategy when writing words they were unsure how to spell (see Appendix for protocol). First, participants were encouraged to generate plausible spellings and asked to evaluate the spelling on the basis of residual orthographic knowledge ("Does it look right?"). They were then encouraged to use a sublexical strategy to "sound out" any parts of the word they were unsure about, and again asked to check the spelling against their lexical knowledge. If still unsure, or if they wanted to check their spelling for accuracy, the final step was to enter the word into an electronic spell-checking device, the Franklin Speaking Language Master (Franklin Electronic Publishers, Burlington, NJ). This particular device is sensitive to both form-related and phonologically plausible errors. Misspelled words generate a list of possible correct lexical options; a "say" button allowed participants to hear selected words to check for accuracy.

During treatment sessions, low-frequency, irregularly spelled words were presented for writing-to-dictation. Participants were trained to follow the steps to derive correct spellings. When they demonstrated mastery of the problemsolving approach for single words, they were trained to use the strategy for spelling words in sentences: first, writing sentences to dictation and progressing to self-generated sentences using pre-selected low-frequency words. Daily homework consisted of spelling low-frequency words presented auditorily using a recordable photo album. Homework sheets were provided that were structured to guide participants to document the steps used to resolve spelling difficulties. Homework was reviewed at each treatment session to check for accuracy and to maintain accountability. As participants progressed, they were required to write sentences to dictation, and to generate sentences for homework. They eventually progressed to self-generation of written paragraphs on predetermined topics, using the problemsolving approach for spelling as needed.

#### Outcome Measures

Pre- and posttreatment spelling accuracy on the ABRS were compared by item using the McNemar test, providing an indication of generalized improvement. Participants were also given the opportunity to use the spell-checker to evaluate and self-correct spelling; hence, posttreatment scores included responses with and without the spell checker.

## **Results**

All three participants completed oral reading treatment concurrent with interactive spelling treatment. Treatment outcomes are shown in Table 2 and Figures 3 to 6.

#### $PI$

P1 received 12 sessions of treatment over a period of 6 weeks. Passages corresponding to reading levels of Grades 4.5 to 6 were used to probe reading performance and for repeated oral reading. P1's reading rate on trained



Table 2. Average pre- and posttreatment reading performance on trained and novel probe passages, and alternate forms of the Gray Oral Reading Test–3 (GORT–3).

passages improved from an average of 67.8 wpm (range =  $33-102$ ) at first reading to 85 wpm (range = 54–108), which was a significant increase,  $t(4) = 3.40$ ,  $p = .014$  (Figure 3a). P1 also demonstrated a significant increase in reading rate for the GORT–3, improving from an average of 85.6 wpm to 103.3 wpm,  $t(10) = 7.02$ ,  $p < .001$  (Figure 3b). Reading rate and accuracy for novel probe passages also improved (Table 2). Single-word reading was significantly faster posttreatment, decreasing from an average of 1,365 to 1,210 milliseconds,  $t(144) = 4.12$ ,  $p < .001$ . P1's average reading reaction time posttreatment was similar for four-, five-, and six-letter words, but seven-letter words required an additional 300 milliseconds (Figure 3c). The word-length effect was no longer significant,  $r = .774$ ,  $p = .113$ . P1's few residual reading errors tended to be visually similar to the target, for example, change for charge; she no longer made errors that resulted from using a sublexical phonological strategy for reading.

Improved spelling accuracy on untrained words was evident by comparing ABRS scores before and after treatment, McNemar  $\chi^2$  (1) = 6.13, p = .013. Spelling of irregular words improved by 12.5% and most residual errors were self-corrected using the spell-checking device (Figure 3d). Residual spelling errors were all phonologically plausible (e.g., QUIRE for choir).

## $P<sub>2</sub>$

As P2 was nearing 6 weeks of treatment, he experienced a fall and was subsequently hospitalized due to a small subdural hematoma. When he returned to treatment 2 months later, readministration of the Raven's Coloured Progressive Matrices (Raven, Court, & Raven, 1990), the BNT, and the reading/spelling battery revealed performance that was comparable to that before his fall. As he did not appear to have any lasting cognitive effects resulting from the fall, the decision was made to continue treatment. Another 24 sessions were implemented, so that ultimately, P2 received 32 sessions of treatment over a 6-month period. This was a longer treatment duration than originally planned. However, it seemed warranted in response to the treatment disruption, and due to potential limitations related to his visual field impairment. Probe materials were selections from the SRA Reading Lab series at 2.0 grade level, and practice texts comprised passages at approximately 2.0 to 3.0 grade levels. Reading rates for practice texts increased from an average of 67 wpm (range  $=$  58–85) at first reading to an average of 90 wpm (range  $= 81-95$ ) at last reading (Figure 4a), a significant difference,  $t(16) = 9.88$ ,  $p < .001$ . P2 also demonstrated faster and slightly more accurate reading of novel passages over the course of treatment (Table 2). Text reading rate on the GORT–3 also improved significantly from 56.5 to 69.3 wpm,  $t(7) = 3.00, p = .010$ (Figure 4b). His reading of single words was faster after treatment, with average reaction times decreasing from 1,808 to 1,511 milliseconds,  $t(143) = 4.73$ ,  $p < .001$ , but the influence of word length was now significant,  $r = .974$ ,  $p = .013$  (Figure 4c). P2 showed a shift toward more visually similar word errors, but still made some phonologically plausible errors in oral reading.

Figure 3. Treatment results for participant 1 (P1). (a) Reading rate on practiced passages; (b) reading rate and number of errors on untrained GORT–3 passages. (c) Pre- and posttreatment reading reaction times; (d) pre- and posttreatment reading/spelling accuracy. \*Significant increases in text-reading rate ( $p < .001$ ), single-word reading rate ( $p < .001$ ), and overall spelling accuracy posttreatment ( $p < .05$ ).



b. Pre-post reading rate (words/minute) and accuracy c. Pre-post reading reaction time by word length. (# errors/100 words). Rate\* $p < .001$  $RT^*p < .001$ , WLE $\bullet$  -  $\bullet$  =  $p < .05$ 





P2's overall spelling accuracy improved significantly following treatment, McNemar  $\chi^2$  (1) = 8.1, p = .004, demonstrating generalization of spelling improvement to untrained words (Figure 4d). Irregular word spelling improved significantly from 47.5% to 65% posttreatment, McNemar  $\chi^2$  (1) = 5.14, p = .023. Despite his improvements, it was

apparent that P2 had residual impairment of orthographic knowledge, as he relied on phonological decoding to read and spell some words. For spelling, P2 learned to selfdetect and correct most of his errors using the electronic spelling device, so that he made dramatic functional improvements.

Figure 4. Treatment results for participant 2 (P2). (a) Reading rate on practiced passages; (b) reading rate and number of errors on untrained GORT–3 passages. (c) Pre- and posttreatment reading reaction times; (d) pre- and posttreatment reading/spelling accuracy. \*Significant increases in text-reading rate ( $p < .05$ ), single-word reading rate ( $p < .01$ ), and irregular word spelling accuracy posttreatment ( $p < .000$ ).



b. Pre-post reading rate (words/minute) and accuracy (# errors/100 words). Rate\* $p < .01$ 





d. Reading/Spelling accuracy.  ${}^*p = 0.023$ 



## $P<sub>3</sub>$

P3 received 12 sessions of treatment over a 6-week period. Text materials were selections from the SRA Reading Lab series at approximately Grade 2.5 to 3 levels of difficulty. Reading rates for practice texts increased from an average of 49 wpm (range  $= 35-73$ ) at first reading to an average of 72 wpm (range  $= 61-98$ ) at the last reading

(Figure 5a), a significant difference,  $t(8) = 2.39$ ,  $p = .022$ . Reading rate and accuracy for novel probe passages was variable, as demonstrated by small effect sizes (Table 2), but average text reading rate on the GORT–3 increased significantly from 52.8 to 70.2 wpm,  $t(9) = 3.30, p = .005$  (Figure 5b). P3 demonstrated a significant improvement in single-word reading latency, from an average of 2,933 to 1,697 milliseconds Figure 5. Treatment results for participant 3 (P3). (a) Reading rate on practiced passages; (b) reading rate and number of errors on untrained GORT–3 passages. (c) Pre- and posttreatment reading reaction times; (d) pre- and posttreatment reading/spelling accuracy. \*Significant increases in text-reading rate ( $p < .01$ ) and single-word reading rate ( $p < .000$ ) posttreatment.







c. Pre-post reading reaction time by word length. RT\*p < .001, WLE $\bullet$  -  $\bullet$  = p < .05







Figure 6. Results of posttreatment self-rating questionnaire completed by the three participants with alexia and agraphia.



(Figure 5c),  $t(123) = 9.57$ ,  $p < .001$ , and the word-length effect was no longer significant,  $r = .796$ ,  $p = .102$ . His text reading was improved but still slow at about 1 second per word, with residual reading errors visually similar to the target.

P3's overall spelling accuracy improved significantly from 73.8% to 83.8% after treatment, McNemar  $\chi^2$  (1) = 6.13,  $p = .013$ , demonstrating improved spelling of untrained words. Irregular word spelling improved by 15%, but P3 had some residual difficulty self-detecting errors such that irregular word spelling was still less accurate than regular words (Figure 5d). His residual spelling errors reflected use of a phonological strategy at least half of the time.

#### Participant Evaluation of Treatment

After treatment was completed, the participants were interviewed and completed a survey to assess their perception of reading and spelling abilities after treatment. As shown in Figure 6, a 7-point rating scale was used to respond to four questions. All three also reported better reading and spelling after treatment, and improved overall confidence in their written language skills.

#### **Discussion**

In this study, three individuals with damage to lvOT due to left PCA stroke demonstrated written language impairments characterized by a combination of peripheral and central orthographic processing deficits. Prior to treatment, all three participants had slow reading rates and an abnormal word-length effect typical of LBL reading, suggesting a disruption in the parallel visual processing of letter strings. In addition, they showed a regularity effect in spelling and an overreliance on a sublexical strategy that resulted in a preponderance of phonologically plausible errors (surface agraphia), with two individuals also showing surface alexia. All three responded well to the behavioral treatment protocol designed to address acquired reading and spelling deficits concurrently. Treatment outcomes included faster reading reaction times for single words and improved reading rate for text, as well as improved spelling accuracy for irregular words that were not specifically trained. The positive outcomes from the reading treatment (MOR) and interactive spelling treatment were consistent with previous

case reports when the treatments were administered in isolation (Beeson, 1998; Beeson et al., 2000, 2005, 2010; Moyer, 1979; Tuomainen & Laine, 1991), but this study demonstrated an efficient concurrent treatment regimen.

As portrayed in Figure 1b, the goal of MOR treatment is to shift the reader away from the abnormal LBL strategy toward a lexical (whole-word) approach used by skilled readers. Posttreatment reading reaction times showed that all participants were faster at reading single words, and there was evidence of shifts toward a whole-word approach in P1 and P3, who showed resolution of the word length effect. In the case of P1, she had similar reading reaction times for four-, five-, and six-letter words after treatment, suggesting that she improved her ability to apprehend letter strings in parallel up to about six letters, whereas sevenletter words still required more time. P3, who was the slowest reader at the outset (requiring about 2–3 seconds/word), showed the most obvious shift from a serial LBL reading approach to a more parallel process wherein reading reaction time was similar across word lengths. P2 read single words faster after treatment, but the word length effect was significant, suggesting reliance on serial processing of words presented in isolation.

With regard to text reading, all three participants increased reading rate on practiced text an average of 21 wpm  $(range = 23-17.2)$ , and more importantly, they improved reading rate on the GORT–3 by an average of 16 wpm  $(range = 12.8-17.7)$ . These gains in reading of new text (i.e., an alternate form of the GORT–3) show impressive generalization. Consistent with these data, participants judged their reading to be "better" (P2) or "a lot better" (P1 and P3) after treatment. As expected, reading rate for text was faster than for individual words, suggesting the benefit of semantic and syntactic context for word recognition. That is not to say that improvement was based entirely on top–down processing because single-word reading in isolation was faster as well, and the word-length effect had resolved in P1 and P2.

Spelling treatment implemented concurrent with MOR treatment was intended to promote the interactive use of lexical and sublexical information to provide a means for self-cuing and self-correction of spelling errors. We expected that engaging in this effortful problem-solving process using preserved sublexical processing skills and a compensatory aide (spell checker) would also strengthen

degraded orthographic representations. The generalized improvement in spelling of untrained words indicated that treatment had the desired effect. Similar to previously reported cases (Beeson et al., 2000, 2010), the effort devoted to retraining self-detection and correction of spelling errors using the electronic spell checker did not simply provide a compensatory strategy for weak spelling skills, but also had a rehabilitative effect. Interactive spelling treatment strengthened orthographic representations, as evidenced by improved spelling of irregular words and a reduction in the regularity effect in spelling.

Both treatments were intended to strengthen and promote lexical processing of orthography: to support word recognition in reading and retrieval of word-specific orthographic knowledge in writing. Writing inherently engages reading as one reviews and evaluates written output, and as orthographic representations become stronger, greater bottom–up support is available for both spelling and reading. The interactive spelling treatment is, by design, a deliberate and somewhat slow process, whereas MOR treatment is intended to push for more rapid word identification. Thus, the concurrent treatment was intended to maximize the synergistic effects of bottom–up processing, while capitalizing on the top–down benefits of MOR to facilitate more rapid lexical processing. The contribution of both treatments to strengthening orthographic representations is depicted in Figure 1b.

Although we combined treatments in order to effect maximal benefit in this study, additional research is warranted to compare the singular or sequential effects of MOR and interactive spelling treatment to the combined approach used in this study. One comparison of interest can be made to Participant RB in Beeson et al. (2005), whose profile was similar to the participants in this study in that his LBL reading was accompanied by a significant regularity effect in spelling (i.e., surface agraphia). RB received MOR treatment only, and the outcomes included faster text reading (by about 19 wpm) and resolution of the word-length effect, but there was no significant improvement in spelling and the regularity effect persisted. This suggests that MOR treatment was specific to reading and did not strengthen orthographic representations to the extent that it benefitted spelling.

We note that the participants in this study were quite similar in their pretreatment profiles and positive response to treatment. Although the integrity of their perceptual visual fields differed, that did not prevent a good treatment response. It is likely, however, that a visual field impairment could limit the ultimate degree of recovery. Recent research examining the effects of hemianopia on reading latencies have shown that right hemianopia is likely to disrupt parallel processing of letter strings along the order of 50 to 160 milliseconds per letter (Leff et al., 2001; Sheldon, Abegg, Sekunova, & Barton, 2012). Our participants with visual field cuts (P1 with right hemianopia, and P2 with right hemianopia plus a left lower quadrantanopia) improved their single-word reading reaction times after treatment, but they were still averaging over 1,000 milliseconds per word.

After treatment, the residual word length effect for P1 was only notable for seven-letter words. However, P2 had a residual word-length effect that averaged about 172 milliseconds per letter, which approaches a latency that could be accounted for by his marked visual field defect, rather than an LBL reading approach.

Time post onset of brain damage was also not a limiting factor in the response to treatment. P1 was of particular interest because we had the opportunity to document aspects of "natural" recovery that occurred between 2 weeks poststroke to the beginning of treatment at 8 months post. At 2 weeks poststroke, her profile included surface alexia, characterized by a pronounced regularity effect and the production of phonologically plausible reading errors that were in fact nonsense words, such as blude for blood. At that time, P1 was aware of the absurdity of some of these responses and chuckled at them, so it was interesting to note that by 4 months poststroke, she produced few phonologically plausible reading errors. Instead, her errors tended toward visually similar words to target words, suggesting an inclination to move away from using a phonological strategy that produced nonwords, in favor of a "guessing" approach to identify letter strings as real words. This awareness that a phonological approach to reading produces nonwords may explain why surface alexia is infrequently observed in chronic stroke patients with intact semantics. This is in contrast to individuals with persistent surface alexia, such as those with semantic dementia, who presumably do not recognize nonword errors due to eroding semantic knowledge (cf. Woollams, Ralph, Plaut, & Patterson, 2007). In contrast to reading, phonologically plausible spelling errors are functional and relatively well tolerated, even in the writing of healthy adults, which may underlie the retention of a phonological strategy for spelling, but not reading. This would suggest that recovery from damage to orthographic knowledge (in the face of relatively preserved semantics) is likely to evolve to persistent surface agraphia with mild or absent surface alexia.

Although P1 and P3 initiated treatment within the first year poststroke, P2 was 9 years post onset of alexia and agraphia when treatment was implemented. His response to treatment was equally robust in comparison to the other two. This is confirmatory that the changes observed in response to treatment are not likely to reflect a continuation of natural recovery in these participants. Indeed, P2's improvement is all the more noteworthy in light of the fact that similar cases of individuals who demonstrated positive response to MOR treatment were within 2 years postonset of impairment (Beeson et al., 2005; Moyer, 1979; Tuomainen & Laine, 1991).

Regarding the duration of treatment for a given individual, we planned to administer 6 weeks of treatment because our previous research indicated that amount of time was adequate to complete the interactive spelling treatment protocol. The 6-week treatment proved to be a good duration for P1, who improved her reading rate such that she was reading for pleasure again. Her spelling also improved to within the range of age-matched controls. She expressed

satisfaction with the treatment outcomes, and we also considered that her return to pleasure reading might promote continued improvement without direct treatment, as observed in HL reported by Beeson (1998). P3 also received 6 weeks of treatment and was pleased with the outcomes. We note, however, that he continued to have a slow reading rate and residual spelling deficits that may have benefited from additional treatment directed toward further strengthening of orthographic representations and wholeword reading strategies.

As noted, P2 received more treatment sessions than P1 and P3 in response to the interruption of his treatment, and also due to concern about the limitations posed by his visual impairment. A review of his treatment data showed that after 6 weeks of treatment, P2 demonstrated improved reading rate for novel text (effect size  $d = 1.34$ ), and ultimately the treatment effect size increased to 2.00. In other words, he benefitted from the additional 24 sessions of treatment after he returned. This highlights the fact that the duration of treatment may best be determined on an individual basis. A research context may require testing at fixed intervals in order to discern the trajectory of change, but in a clinical setting, discharge from treatment may be based on a number of factors including functional goals and cost–benefit assessment for time and money. Future studies manipulating treatment duration and/or intensity could further elucidate this issue.

In summary, the positive treatment outcomes from this study support the combined use of MOR along with interactive spelling treatment for individuals with alexia and agraphia associated with lvOT damage. Despite the limitations of our small sample size and some heterogeneity, all three participants responded favorably to this concurrent treatment approach. Our findings highlight the need to address both written language modalities, and future studies may further explore the relative contributions of each treatment, as well as the dynamic interaction of the processes involved.

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#### Appendix

Protocol for Multiple Oral Re-Reading and Spelling Treatment

