Whole-word training of high-frequency words in a case of pure alexia

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The final copy of this thesis has been examined by the signatories, and we Find that both the content and the form meet acceptable presentation standards

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ABSTRACT

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Pure alexia is an acquired reading disorder, in the absence of a more pervasive language disorder. People with pure alexia are unable to read whole words and therefore employ a strategy of reading each word one letter at a time. Treatments for pure alexia attempt to improve the patient's reading ability by 1) training letter-by-letter reading skills or 2) training the patient to recognize whole words. The current study attempted to assess the efficacy of a treatment that trained explicit whole-word recognition of the 20 most frequently used words in written English. The participant, EF, had severe pure alexia and used a letter-by-letter reading only treatment phases, and a post-treatment assessment phase. During treatment, the 20 target words were repeatedly presented to the participant for less than one second in duration, using an Android tablet. Treatment efficacy was determined by how well the participant learned the trained words and how much this learning influenced functional reading ability. Results of the study indicated that there was no significant change in EF's ability to read the trained high-frequency words. Likewise, no improvement was seen in his general reading ability. Possible reasons that no improvement was shown include clinician error and flaws in the design of the study. Another possibility is that the participant, EF, was not the most suitable candidate for the treatment. More research of treatments for moderate to severe pure alexia is needed.

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CHAPTER 1

INTRODUCTION

Brain injury may potentially cause a variety of different types of expressive and receptive language problems. An acquired impairment in reading, known as alexia, is common in brain injury, and may occur in the presence or the absence of more pervasive language impairments. One of the more interesting types of reading impairments is pure alexia, a condition that is not associated with a more general aphasia or with difficulties in written output.

Pure alexia is characterized by an inability to read words as whole units. People with pure alexia often compensate for this deficit by reading words one letter at a time. For this reason, pure alexia is also known as "letter-by-letter" reading. As a result of this letter-by-letter reading strategy, a clear length effect can be shown in people with pure alexia. That is, words that are longer in length take longer to read. In addition, words with more letters are more likely to be misread (Weekes, 1997). Despite the inability to access orthographic word forms when presented with visual input (letters), people with pure alexia do recognize orally spelled words and write letters, words, and sentences (Friedman & Lott 2000).

Pure alexia is typically the result of damage to the occipitotemporal area of the left hemisphere. The left fusiform gyrus is particularly sensitive to visual word forms. This area, which may also be called the visual word form area (McCandiss et al, 2003), is responsible for the perception of visual word forms, a skill necessary for rapid reading. Lesions in the visual word form area can prevent the patient from transferring visual input from the visual cortex in the right hemisphere to the language processing areas found in the left hemispheres. In other words, even though language processing is intact, there is an impaired ability to transfer visual information from printed words to the necessary areas of the brain to be processed as language input (Friedman & Lott, 2000).

CHAPTER 2

LITERATURE REVIEW

Historically, the numerous theories attempting to explain the deficits seen in pure alexia can be divided into two main classes. One class of theories suggests that the deficit occurs early on during activation of the orthographic representation of words. Supporters of this view point to the fact that people with pure alexia typically have an impairment in letter recognition (e.g., slower and less accurate). These theories can be classified as "peripheral" theories¹ (Behrmann, Plaut, & Nelson, 1998).

A second class of theories proposes that deficits occur later in the reading process. These theories can be classified as "central" theories (Behrmann, Plaut, & Nelson, 1998). Proponents of central theories stress that patients with pure alexia are able to access lexical and semantic information even when they are unable to explicitly read a word. This is typically shown by asking patients to perform lexical decision tasks and semantic classification of words (e.g. living vs. non-living) but only presenting words for a very brief time period so that the patients are discouraged from reading the word one letter at a time. Coslett et al. (1989) hypothesized that people with pure alexia are able to perform at better than chance on these tasks because, even though visual orthographic information processed in the right hemisphere is unable to transfer adequately to the left hemisphere for explicit whole-word reading, word information can still be processed by a primitive semantic system (analogous to the visual word form area) in the right hemisphere.

Treatment approaches for pure alexia fall into two main categories, corresponding to these two main theories of the disorder. Treatments that follow the peripheral model aim to

¹ The term peripheral, in this context, is not implicating the eye itself or its connections to the primary visual cortices.

improve impaired letter processing by training letter-by-letter reading abilities. Such treatments have been shown to effectively increase rate of letter recognition and speed of reading (Seki, Yajima, & Sugishita, 1995; Lott, Friedman, & Linebaugh, 1994; Lott & Friedman, 1999). These treatments often include tactile or kinesthetic reading. In this approach, the patient may trace or copy the outline of each letter in a word (Seki, Yajima, & Sugishita, 1995), trace letters into the palm of their hands (Lott, Friedman, & Linebaugh, 1994), have letters traced in their palm (Sage, 2005), etc.

The primary benefit of training letter-by-letter reading is that the strategy can be generalized to untrained words, increasing the speed of reading on both trained and untrained tasks (Lott & Friedman, 1999). However, even when letter-by-letter reading improves, reading rate is still subject to a length effect and reading is still slow and laborious compared to a typical reader (Sage et al., 2005).

For this reason, many treatments follow the central theoretical model. These treatments tend to use semantic categorization tasks and lexical decision tasks (e.g. "Is this word an animal or a vehicle?") (Gonzalez-Rothi, 1998; Gonzalez-Rothi & Moss, 1992) or train whole words using a brief presentation of the target words (Friedman & Lott, 2000; Behrmann & McLeod, 1995). While these approaches have been shown to improve reading ability for trained stimuli, gains do not generalize to novel words. Because whole-word training does not generalize to untrained words, a whole-word reading technique may not be an effective strategy unless used in conjunction with a letter-by-reading strategy for untrained words.

A similar duel approach of training both speed and accuracy of letter-by-letter reading and whole-word recognition was attempted by Sage et al. (2005). The patient in this study, FD, had severe pure alexia. Before treatment FD utilized a letter-by-letter reading strategy for many words but also demonstrated partial lexical-semantic knowledge of words presented briefly (25 milliseconds). However, in both letter-letter reading and whole-word reading measures, FD made frequent errors.

FD's treatment began with whole-word treatment, designed to utilize the patient's implicit semantic knowledge of whole words. The second treatment approach trained letter-by-letter reading to increase the speech and accuracy of reading.

During the seven weeks of whole-word training, the patient's family showed him word cards and then told him the word. FD would then repeat the word five times. After four weeks the patient identified 16 of the 30 words (53%) which was below the criterion for success (80% correct within 30 seconds). During the following three weeks a word shape description strategy was added to the treatment. Attention was called to the visual features of the word as well as reading the word out loud. The patient scored 17 of 30 words after the three weeks.

During letter-by-letter therapy a hierarchal approach was used. First, family members would present word cards to FD and read the words aloud, one letter at a time, while tracing letters in FD's palm. FD would then repeat the word letter-by-letter. When FD could successfully say the word, he was asked to read aloud the letters himself and then say the word.

In both treatment approaches, the patient's word reading was measured for accuracy as well as speed. The patient showed improvement after both treatments, particularly to words that were of interest to him. Letter naming accuracy however was resistant to change. FD actually stopped using letter-by-letter reading in favor of a whole word reading strategy. Though he was not able to accurately identify all the words in a passage, FD reported that he was able to understand the basic message of what he was reading. For example, when reading religious magazines that were of personal interest to him, FD was able to gauge the general nature and content of what he was reading.

There are those (Behrmann, Plaut, & Nelson, 1998, Friedman & Lott, 2000) who feel that neither the peripheral theories nor the central theories are adequate for explaining **both** impaired letter processing and the ability to access lexical information. Based on a review of 57 studies on pure alexia, Behrmann, Plaut, & Nelson (1998) proposed a third major theory of pure alexia that suggests that reading difficulties arise from a perceptual impairment of word recognition. This impairment allows only partial parallel (simultaneous) recognition of the letters in a word. Because the word activation is too weak for the patient to explicitly identify the word, they must resort to reading each individual letter. However, the initial parallel activation of the word, though weak, is able to support a partial lexical activation. This explains the ability that many patients have to perform at better than chance levels in semantic categorization and lexical decision tasks.

Behrmann, Plaut, & Nelson (1998) also proposed that the partial lexical activation of the word feeds back to the letter level to facilitate letter recognition. For evidence of this "top-down" feedback, the authors state that there is an imageability effect as well as a frequency effect on word recognition. That is, words that have a higher frequency of use, and words that are more imageable are more quickly and/or more accurately recognized. In order to reach this conclusion, the authors reviewed 26 cases in which frequency was tested. Frequency influenced word recognition in 23 of the 26 cases. Additionally, out of 19 subjects that were tested for imageability, 12 showed that imageability positively influenced word recognition.

A similar model of reading aloud (see figure 1), proposed by Friedman and Lott (2000), begins at the level of letter recognition. At this level, letters are recognized either "automatically and in parallel" or "explicitly and serially". This information activates orthographic representations at the level of "orthographic lexicon" and then diverges into "phonological lexicon" in the left hemisphere and "semantic lexicon" in the right. Information is transferred from the semantic lexicon to the phonological lexicon and then produced as spoken language. In pure alexia (letter-by-letter reading), the letter identification system fails to recognize letters automatically and reverts to explicit, serial reading of each letter.



Figure 1. Model of oral reading proposed by Friedman and Lott (2000)

Using this model as a theoretical framework, Friedman and Lott (2000) explored whether or not their patient with pure alexia could be trained to read whole words by accessing the right hemisphere semantic reading route. The patient treated in this study, a 46-year-old, left-handed man identified as RS, presented with mild pure alexia after the removal of a hemangiopericytoma in the left occipital lobe.

Several different treatment methods were analyzed in this study. In the first of these, RS was asked to make semantic judgments about words rather than using letter-by-letter reading. At

the beginning of each set of words, the patient was told what the target category was and instructed to answer "yes" if the word fit the category and "no" if the word did not. For example, he was presented with a list that contained both bird names and orthographically similar distractor words. He was then asked to identify if each word was a bird or not a bird. The participant received treatment for 16 weeks, two times per day. Improvement was seen in trained words and length effects decreased substantially. However, no significant gains were made in untrained words.

In subsequent experiments, the researchers switched from categorization of words to oral reading of words. Twenty-word lists were presented in categories. Ten of the words were from the category while the other 10 were distractor words that were matched to each category word based on similar orthography, length, frequency of use, and part of speech (all nouns). At the beginning of each set of words, the participant was told what the target category was and instructed to read each word in the set aloud. Words were presented on a computer screen for a duration of 30 milliseconds. While 30 milliseconds is sufficient time for typical readers to recognize a word, it is a short enough time period that the patient was unable to utilize letter-by-letter reading. RS read aloud each word and auditory feedback regarding accuracy was immediately provided. Training lasted 22 weeks with a frequency of three times per week. RS reached 90% accuracy on trained words, each set of words taking five to ten weeks to achieve maximum treatment benefit.

The oral reading of nouns, though not a strictly semantic task since the patient was not asked to identify semantic categories, still utilized semantically rich nouns that had been divided by semantic category. In order to test if RS's success in this task was semantically based, the researchers examined the learning of functors (function words such as prepositions or pronouns). Training of functors was administered three times a week. With three weeks of training, RS reached 90% criterion reading ten trained words, while performance with untrained words did not change significantly. Pseudowords, which contain no semantic value, were also trained in the same manner and showed some improvement, but never reached criterion level.

Friedman and Lott (2000) hypothesized that the oral reading of words (nouns as well as functors) improved as much as semantic categorization tasks because the gains were occurring somewhere **before** the level of the orthographic lexicon. Orthographic units were only weakly activated after receiving visual input. Therefore, the repeated presentation of written words with immediate feedback was able to strengthen the link between letter recognition and the orthographic representations.

The authors concluded that in pure alexia, a deficit in the "rapid parallel identification of letters" is what causes patients with pure alexia to read letter-by-letter. A "semantic effect" is seen when serial letter recognition only weakly activates the orthographic lexicon, resulting in partial transference of semantic information that is insufficient for phonological activation.

This study also has implications for the importance of word frequency in pure alexia treatment. Because the trained function words had a much higher frequency of use than the more semantically rich nouns (and of course, the pseudowords), the fact that functors were learned more readily suggests that improvement may have been related to written word frequency. This could be due to a stronger connection between letter identification and orthographic representation in words that are used more frequently.

In a recent study by Lacey et al. (2010), the authors examined gains made by four participants after eight weeks of therapy using the Multiple Oral Re-reading (MOR) treatment tool. MOR involves reading text out loud multiple times a day and has been shown to improve speed of reading on trained as well as untrained passages. However, Lacey et al. found that gains were only "generalized" when the novel text contained a critical mass of the words contained in the trained passages. It was proposed that the success of the alexia treatment Multiple Oral Re-reading (MOR) may be due primarily to the training of high frequency functor words, not improvement in top-down processing.

Training the most frequently used words in written English may have the greatest impact on functional reading because they appear more often than any other words. From the research (Lacey et al., 2010; Friedman & Lott, 2000; Behrmann, Plaut, & Nelson, 1998), it would also seem high-frequency words may also be the easiest to train since they occur more often in written language, creating for readers a stronger connection between the words and their orthographic representation.

As discussed, Friedman and Lott (2000) demonstrated that a patient with pure alexia, RS, could learn to identify whole words through repeated brief presentation and immediate performance feedback. However, learning only applied to trained words. Thus, for this approach to make a significant difference in functional reading, high frequency words must be trained. Friedman and Lott suggest the following:

Are we to retrain all words of the language? In fact, if one considers that a large proportion of most sentences is composed of a small number of very high frequency words, the task seems far more manageable. If patients could be trained to rapidly recognize the 125 or 150 most frequent words of the language, it is likely that overall reading could improve substantially. (p. 236)

In one case study reported by Friedman and Lott (1997), the patient, FT, was trained in whole-word reading using repeated brief presentations –approximately 30 milliseconds– of very

common words. This phase of treatment was referred to as "tachistoscopic word recognition" and was accompanied by a second treatment phase which trained letter-by-letter reading. The goal of combining these two forms of treatment was to allow for the patient to maximize reading efficiency. Because whole word reading treatments have not shown generalization to non-target words, the target words used for treatment were the 120 most commonly used English words. Ideally, the patient would improve in her ability to rapidly read common words using a whole-word reading strategy and then switch to using a speeded letter-by-letter reading strategy for non-trained or less common words.

Results of the tachistoscopic phase showed improved speed when reading sentences with only target words, and a much less pronounced improvement when reading sentences with both target and non-target words. These results suggest that the benefits of whole-word training were, as predicted, specific to the targeted stimuli. In contrast, the letter-by-letter reading strategy taught in stage two of treatment was predicted to generalize to all words. The results of the second phase were not published.

CHAPTER 3

PURPOSE

The purpose of the current study was to investigate the efficacy of training whole-word recognition of high-frequency whole words in a patient with pure alexia. Treatment focused on the training of 20 of the most commonly used in newspapers and magazines. Although these core vocabulary words are among the most frequently used across all media and genre, we chose to create the list based specifically on newspaper and magazine corpora because these are the media that the participant is most interested in reading.

The goal of the treatment is to improve the participant's functional reading fluency (speed and accuracy of oral reading). The participant, EF, participated in an intensive four-week program that trained the 20 most common words in written American English. During this time, we attempted to answer the questions:

- Does training whole-word recognition of high-frequency words improve reading of the trained words?
- 2. Does training whole-word recognition of high-frequency words improve functional reading ability?
- 3. Does training whole-word reading ability generalize to untrained words?
- 4. Are changes in performance maintained after training has finished?

CHAPTER 4

METHOD

Treatment Design

This study was a single-subject, AB₁B₂A design that consisted of a pre-treatment assessment, two sequential treatment phases, and a post-treatment assessment. These different phases will be referred to in this report as the Pre-Treatment Phase, Treatment Phase I, Treatment Phase II, and Post-Treatment Phase. One set of stimuli words was treated in each of the treatment phases while the other list was untreated, allowing for measurement of both treatment generalization and maintenance.

Participant

EF was a 66-year-old left handed male who suffered a left middle cerebral artery cerebral vascular accident (CVA) in 2004. He was eight years post-onset at the time of this study. Immediately following his CVA and subsequent 10-day coma, EF was described as presenting with aphasia, dysarthria, right hemiparesis, and vision problems. Prior to his CVA, EF was the founder of a financial company and was working at the time of his stroke. He programmed software, traveled often to assist clients, and engaged in public speaking. He was also an avid reader. Before founding a financial company, EF attended several years of college but left before earning his degree.

For several months during the first year post onset, the participant received speech language services that focused on recovery of speech, language, and memory. Treatment was discontinued approximately eight months post onset. EF did not receive treatment for the next several years until he was evaluated at the University of Colorado at Boulder Speech Language and Hearing Center (SLHC) in 2012, approximately seven years post-onset. EF's primary reported concern at this time was his inability to read. Both formal and informal evaluation of his reading, writing, language, cognition, and Tspeech was conducted. Language measures administered included the Western Aphasia Battery (Kertesz, 1982), the Boston Naming Test (Kaplan, Goodglass, & Weintraub, 2001), and the Psycholinguistic Assessment of Language Processing in Aphasia (Kay, Lesser, & Coltheart, 1992). The results of these measurements are summarized in Table 1.

Table 1

Boston Naming Test (BNT)			
<u>Test</u>	Score		
Boston Naming Test	43/60		
Psychological Assessment of Lang	uage in Aphasia (PALPA)		
Subtests	Score		
18. Mirror Reversal	18/18		
19. Upper Case-Lower Case Matching	25/26		
20. Lower Case-Upper Case Matching	25/26		
22. Letter Naming and Sounding	23/26		
23. Spoken-Written Letter Matching	19/26		
29. Letter Reading (3 letter words)	5/6		
29. Letter Reading (4 letter words)	0/6		
Western Aphasia Battery-R Part 1			
Subtests	Score		
Spontaneous Speech Score	20/20		
Auditory Verbal Comprehension	9.8/10		

Appendix of scores from EF's evaluation in 2012

Repetition	9.6/10
Naming and Word Finding	8/10
Aphasia Quotient (AQ)	94.8

According to results of informal assessment and the Western Aphasia Battery (see Table 1), EF's spontaneous speech was fluent, his verbal comprehension intact, and repetition ability was within normal limits. The Aphasia Quotient of 94.8 indicates that EF was not aphasic at the time of the assessment. In addition, he produced no paraphasias during the assessment. EF's articulation was slow and imprecise secondary to mild dysarthria.

On reading tasks (see PALPA scores Table 1), EF was able to correctly identify which letters were in correct orientation vs. mirror-reversed, as well as to match upper and lower case letters. However, he was only able to identify the letter name or indicate the sound associated with the written grapheme 50% of the time. This was clearly not due to word-finding difficulties as he was also able to accurately and efficiently identify letters and short words that were traced in his palm by the clinician. EF could also correctly name written numbers and mathematical symbols.

On an informal writing task, EF wrote with his premorbid dominant left hand. He wrote his name, address, birth date, and telephone number with no errors. He was also able to spell words aloud and identify words spelled to him by a clinician.

While EF presented with visual problems, informal assessment of vision suggested that EF's acuity is adequate for letters presented in larger than 12 pt. fonts. He also had a visual field cut and a lower quadrant field cut. Therefore, visual stimuli were presented to the upper right visual quadrant and EF was instructed to make any necessary adjustments throughout the treatment study.

Over ten months following his evaluation, EF participated in language treatment at SLHC. EF trained in a number of different skills related to a letter-by-letter reading strategy, including speed and accuracy of letter identification and reading short sentences with auditory feedback. These skills were trained with both upper and lower case letters. EF was able to make modest gains in letter identification ability. However, the most significant gains were seen in the more functional task of reading short phrases.

Stimuli

The stimuli words were comprised of the 20 most frequently used words from newspapers and magazines that are at least two letters long. These words were divided into two lists (see appendix B), List A and List B, and were matched for the length of the words as well as the frequency of use. The words (see appendix A) were selected using the Corpus of Contemporary American English (COCA), a database containing over 450 million words balanced across the genres of fiction, newspaper, magazine, academic, and spoken English. While most frequency lists available in academic literature calculate frequency based on lemma, the COCA allows users to calculate frequency of actual words. For example, the words *be, is, was, are,* and *been* would all be included in the 20 most common words even though they are derived from the same lemma (*be*). The 20 words used in this study were calculated based on the newspaper and magazine corpora and were estimated to comprise approximately 24% of written English (see appendices A and B).

For evaluation purposes, an additional ten untreated words were used during pre- and post-treatment measures, as well as probing tasks during treatment. These ten words were matched to ten of the target words in length and orthography (at least 66% of the letters the same).

The stimuli were presented in all lower case letters and in Arial font, size "Large" (comparable to size 42 in Microsoft Word). Target words were displayed in white letters on a black background using the mobile app Sight Word Flash Cards (van Strien, 2012). The app was displayed using an Android tablet.

Treatment Procedure

The treatment phase was divided into two parts, each lasting 14 days with six sessions per week. Each of the two treatment phases trained 10 of the 20 most frequent words. Three sessions each week were supervised by the primary investigator in order to assure consistency of treatment and allow for ongoing tracking of progress. The other three sessions each week were completed independently at home by the participant. Compliance with the home practice schedule was verified by the participant be marking days on a calendar as was as by verbal report.

During all treatment sessions, stimulus words were presented for 150 milliseconds (ms) each. Prior to the presentation of each word, a visual mask (the + sign) appeared on the screen to fixate EF's gaze. When the participant was ready, he would tap the screen with a stylus and the screen would go blank for 250 ms before presenting the stimulus word for 150 ms.

Each treatment session included two parts. In the first, Part A, the participant was not asked to respond when the stimuli was presented. Instead, the participant was instructed to immediately tap the screen following the visual stimuli. This would activate an auditory presentation of the word produced by the app. The voice for this auditory feedback was a synthesized male voice with a standard American English accent. Each of the ten target words being trained was presented in two sets, each in random order. In Part B of each treatment session, the ten target words were randomly presented seven separate times, totaling 70 word presentations and the participant was asked to orally read each word as it is presented. After EF had responded, he would tap the screen with a stylus and receive feedback via an auditory presentation of the word. Both Part A and Part B together required 40-45 minutes to complete.

Sessions completed with clinician supervision included an additional task, Part C. In this additional task, the participant was asked to choose between two options when presented with each stimulus. For example, the clinician would say, "Is this word *the* or *he*?" before the presentation of the word, *the*, and the participant would respond by saying "the." Part C required 5-10 minutes to complete.

Sessions with the clinician also included the gathering of probe data for both word lists throughout the four weeks of treatment in order to track progress in accuracy of word recognition with a brief presentation. At the beginning of each session with the clinician, all 20 target words and ten untreated words were tested. The probe was the same as Part B of treatment, except that there was no auditory feedback given.

Dependent Measures

Three measures were administered three times during the week before treatment began and during the week after treatment ended. The following is a description of each the three dependent measures.

Dependent measure 1: Accuracy of oral reading with short presentation. Oral reading performance on the 20 target words and ten untreated words was assessed using the

Android tablet (as described above). Stimuli were presented in random order, one time each for 150 ms each². No performance feedback was provided during baseline measurements.

The clinician judged each response to be either correct or incorrect. Correct responses were (1) initiated within a 5 second time period after the stimuli is presented, (2) complete, and (3) accurate. If any phonemes were added, deleted, or changed the response was considered incorrect. The percentage correct was calculated and recorded by a separate clinician.

Dependent measure 2a: Accuracy of word recognition with prolonged presentation.

Oral reading performance of the 20 target words was measured using a PowerPoint slide show on a lap-top computer. The PowerPoint contained the 20 target words and ten untreated words presented in random order. Each word was presented once until EF responded to the stimuli.

To calculate accuracy, the clinician judged each response to be either correct or incorrect. Correct responses were judged to be complete and accurate. If any phonemes were added, deleted, or changed the response was considered incorrect.

Dependent measure 2b: Speed of word recognition with prolonged presentation.

Speed of oral reading performance of the 20 target words was measured along with accuracy, using the same PowerPoint slide. The subject's speed of response was determined by measuring the time between the presentation of the stimuli and the onset of the verbal response.

In order to calculate speed, the clinician used a stopwatch to time the pause between the presentation of each word and the initiation of each response (in milliseconds).

Dependent measure 3a: Accuracy of reading target words in context. A functional reading test was adapted from Dynamic Indicators of Basic Early Literacy Skills, 6th Edition

² In studies of similar treatments, the duration of presentation ranges from 30 ms (Friedman & Lott 2000) to 250 ms (Sage, 2005). Based on observations of the participant's processing speed it was felt that a longer presentation would be most effective.

(DIBELS; Good & Kaminski, 2003). This test was used to test how accurately EF was able to read target words in context. Twelve selections were taken from written stories in the DIBELS, six from a second grade level and six from a fifth grade level (see Appendix C). The reading passages were presented using a PowerPoint presentation similar to that used for dependent measure 2. Videos of this functional reading task were analyzed to determine the percentage of target words that were accurately read.

Dependent Measure 3b: Speed of functional reading. In addition to testing target word accuracy in context, the short paragraph reading task was used to measure how training of target words affected the speed of functional reading. Speed was measured in words-per-minute and was calculated by counting the total number of words in each passage and dividing that number by the number of minutes that EF spent reading the passage.

Data Collection

Performance on all measures was determined using video-recording. While recording, the video camera was aimed toward the computer screen so that the test stimuli could be seen and then compared to the participant's verbal responses. The participant's face was not recorded.

To ensure unbiased data collection and interpretation, the pre- and post-treatment testing sessions were conducted by a clinician other than the principle investigator. This clinician holds clinical certification as a speech-language pathologist from the American Speech-Language-Hearing Association. The primary investigator then received coded video recordings of those sessions, without knowledge of when each session occurred.

During the treatment phase, data was collected by the clinician without the use of a video recording.

CHAPTER 5

RESULTS

The participant attended all planned sessions with the clinician and reported completing all planned sessions at home. Data was collected from three sessions in the Pre-Treatment Phase, six in Treatment Phase I, six in Treatment Phase II, and three sessions in the Post-Treatment Phase. However, data from two sessions of the Post-Treatment Phase and one session of the Pre-Treatment Phase was lost due to clinician error. The remaining data was analyzed in multiple ways to determine the treatment effect, generalization of treatment to untrained words, and maintenance of trained words.

Did reading of the trained words improve?

Improvement in ability to read trained words, or treatment effect, was primarily measured by visually analyzing the data. First, data gathered from Dependent Measure 1 was plotted, as seen in figure 2 and figure 3, and then analyzed to determine the trend for each phase of the study. The participant's scores on this measure reflect how many words he was correctly able to identify when presented with individual words for a very brief period using a tablet device.



Figure 2. Dependent Measure 1, plotted over Pre-Treatment Phase, Treatment Phase I, and Treatment Phase II. This figure represents the number of words correctly identified out of 10 for List A and List B.



Figure 3. Dependent Measure 1, plotted over Treatment Phase I and II of treatment, including trend equations for List A, List B, and the combined total of the two lists. This graph represents the number of words correctly identified out of 10 for List A and B and out of 20 for the "total" of both List A and List B.

From visual analysis of the data, it was apparent that there was no significant treatment effect. In Treatment Phase I, scores measuring the trained stimuli, List A, do not increase. In Treatment Phase II, there is a slight trend upward in scores for List B, the trained stimuli, yet all data points overlap with points in Treatment Phase I. While there also appears to be a slight trend upward in total number of correctly identified words, scores in the last two sessions are approximately equal to scores during the first four sessions. By calculating the trend and correlation (R²), we were able to confirm that, while the trained stimuli for each phase of treatment showed a slight positive trend, neither trend demonstrated strong correlation, and therefore were not statistically significant.

The only potentially meaningful change seen in EF's performance on Dependent Measure 1 was the dramatic increase in accuracy seen between the first and second pre-treatment testing sessions. We suspect that this increase can be attributed to the participant learning the task, rather than any training of the stimuli.

Treatment effect for Dependent Measure 2 was also evaluated primarily through visual analysis. Three separate measures were scored, two pre-treatment and one post-treatment. Scores on this measure reflect EF's performance when asked to read individual words, given an unlimited amount of time to do so. His speed and accuracy on this task are displayed in Figure 4 and Figure 5 below.



Figure 4. Dependent Measure 2a- Accuracy of response



Figure 5. Dependent Measure 2b- Speed of response in seconds

Through visual inspection, we determined that there was no effect of treatment on the speed of response during Dependent Measure 2. In fact, average response time for the words in List A and List B increased post-treatment compared to pre-treatment measures. Inexplicably, the average time for List C words decreased despite the lack of change in accuracy.

The accuracy of response did increases over time, while the untrained stimuli did not. However, the increase between the last pre-treatment session and the post-treatment session was only 10% for both List A and List B, an increase of one word on each list.

In single subject design studies, unless differences between measurements are very large, visual analysis of single-subject treatments can often lead to a Type 1 error (concluding that there is an effect size when there actually is none) (Beeson and Robey, 2006). We therefore determined that other forms of analysis would be helpful in confirming our interpretation of the study results. One alternative to visual inspection is the calculation of effect size to measure change. Effect size is a standard statistical method for determining the existence of a treatment

effect and can be compared across single subject treatments. In addition, effect size allows clinicians and researchers to judge the efficacy of potential treatments Beeson and Robey (2006).

In order to determine effect size, performances on pre- and post-treatment measures are compared to determine effect size using the formula:

$$d_1 = \frac{x_{A_2} - x_{A_1}}{S_{A_1}}$$

 A_2 represents the post-treatment period, A_1 represents the pre-treatment period, \bar{x}_A represents the mean of the data collected in that period, and S_A represents the corresponding standard deviation.

For the most part, studies analyzing pure alexia treatment do not address effect size. Therefore, to interpret effect size, research in the field of acquired language disorders served as a guide for how to calculate and interpret treatment effect size. In a review of 12 studies analyzing aphasia treatment, Robey et al. (1999) calculated that 2.6, 3.9, and 5.8 are small-, medium-, and large-sized effects, respectively. Robey et al.'s (1999) standard was used to judge the effect size found in this study.

Due to lack of data, the increase found in accuracy of word recognition was not analyzed. However, the increase was small enough that we consider it to not represent a significant treatment effect. The effect size for speed of word recognition was analyzed using the method described above. The effect size for both List A and List B were found to be less than 0 (-.32 and -.14 respectively), and therefore did not meet the standard for a small effect size (2.6).

Therefore, our evaluation of treatment effect revealed no significant treatment effect in any of the dependent measures that we analyzed. This conclusion was confirmed using multiple means of analysis, including visual inspection as well the calculation of trend and effect size.

Did functional reading ability improve?

Generalization of treatment to a more functional reading task was measured by comparing pre- and post- performance on Dependent Measure 3. This measure required EF to read short paragraphs that included multiple target words. We measured performance on the task by calculating EF's reading speed in words per second for two pre-treatment sessions and one post-treatment session. We did not analyze reading *accuracy* because EF read all target words accurately when given the context of a short paragraph on both pre-treatment assessments.





The treatment effect for Dependent Measure 3 was determined through visual inspection alone. Post-treatment reading speed decreased compared to the first pre-treatment measurement. No treatment effect was seen on Dependent Measure 3. Therefore, from the data available, there was no generalization of trained skills into a more functional reading task.

Did trained skills generalize to untrained words?

The generalization of treatment to untrained words was determined by visually analyzing EF's performance on List B during Treatment Phase I of treatment. Any improvements seen in the untrained words during the first phase of treatment would potentially suggest that gains from

treatment were generalizing. As can be seen in Figure 2 and Figure 3, there *is* a positive trend seen in List B during Treatment Phase I of treatment. However, it is unlikely that this positive trend signifies generalization. Firstly, a positive trend was not found in the trained stimuli during Treatment Phase I. Secondly, no significant changes were found in EF's speed or accuracy reading List B words when comparing pre- and post-treatment data.

Were changes in performance maintained?

The maintenance of treatment gains was evaluated by visually analyzing and effect size calculations of the performance on List A during Treatment Phase II. If gains from Treatment Phase I were maintained, it is expected that performance on List A during Treatment Phase II would not significantly decrease. As seen in Figure 2 and Figure 3, there is a highly correlated negative trend in accuracy scores for List A during Treatment Phase II. While this may seem to suggest that EF was not maintaining skills, the data from Treatment Phase I indicates that there were no gains in performance and therefore, could not be maintained.

CHAPTER 6

DISCUSSION

Analysis of the treatment data revealed no evidence of significant treatment effect and as would logically follow, no evidence of treatment generalization or maintenance. Faced with the probability that this treatment did not have any significant effect on EF's reading ability, the question we now face is, "Why?" Was the underlying rationale for the current study valid? Was the treatment design suited to answer the treatment questions? Did clinician error during data collection change the results of the study? Was the participant not the most appropriate candidate for this type of treatment? The answer likely lies in a combination of these factors.

Did the study have a valid theoretical rationale?

The current study is based on the available evidence base for pure alexia treatment and current theories of pure alexia. However, as acknowledged in the introduction to this study, there is a lack of consensus as to the underlying processes causing pure alexia. In addition, no randomized controlled studies of pure alexia treatment have been published to date. The literature consists almost exclusively of single subject design studies and case reports. While together these studies may reveal certain patterns in pure alexia and potentially efficacious treatment, they are far from establishing a clear evidence base for treatment. The question of this studies theoretical validity, therefore, cannot be adequately answered at this time.

Was treatment provided in the most effective manner?

While there is little research in alexia treatment on optimum treatment intensity, current literature in aphasia suggests that treatments that involve several hours per week have a greater positive effect than treatments consisting of only 2-3 hours per week (Raymer et al., 2008). For the current treatment, it is possible that an even more intense treatment (e.g. 2 hour-long sessions

per day) would have been more effective in training the target skills. The participant, EF, *was* a very motivated client. However, he frequently expressed feeling fatigued after sessions that were an hour in length. Considering EF's physical condition, it may have been counterproductive to require more than one hour of daily practice.

Studies that evaluated whole-word treatment in patients with relatively mild pure alexia typically trained many more words than were trained in the current study (e.g. 240 total stimuli in Friedman & Lott, 1997). Because EF has moderate to severe pure alexia, we felt that training only 10 target words at one time would be more appropriate. With only 10 words being trained, each treatment session consisted of multiple exposures to a small set of words, allowing EF abundant opportunity for implicit learning of the stimuli to occur. While it may be possible to more successfully train a smaller set of words (e.g. three to four target words), the effectiveness of such a treatment would be questionable, considering the lack of functional benefit for the client.

Did clinician error affect the results of the study?

In order establish the reliability of a measure in single-subject designs, it is necessary to analyze multiple baseline measurements (Hegde, 2003). In the current study, the data from only two baseline measurements were used. In addition, only one set of data from post-treatment sessions was used to determine the effect of treatment. The lack of repeated measures in the posttreatment data makes the post-treatment results more susceptible to threats of reliability. For example, if the participant was feeling ill during that one post-treatment session, the scores may not be an accurate reflection of the participant's actual ability.

While a lack of data may make it difficult to rule out threats to reliability, the available data is sufficient to indicate that the results and subsequent interpretation are valid. The most

compelling argument in favor of this conclusion (that the treatment indeed had no effect on behavior) is the data from dependent measure 1. Dependent measure one was a repeated measure assessed consistently and frequently over time. Therefore, the data from this measure may be considered a reliable measure of EF's behavior over the course of treatment. In addition, being a direct measurement of the behavior being trained, it is the most likely measure to detect changes in behavior. Since there were no significant trends in EF's performance as measured by Dependent Measure 1, it is reasonable to hypothesize that no treatment effect would be seen in other measures. The data from other measures, while incomplete, confirm this hypothesis.

Was EF an appropriate candidate for the current study?

Testing done in March, 2013, 4-5 months prior to the commencement of this study, revealed that EF had impairments in verbal memory (see Table 2). While his non-verbal memory was determined to be relatively strong, EF scored very low on memory tasks that involved language, such as retelling a story after a delay. He also scored poorly in tasks that required explicit learning ability.

Table 2

Test of Memory and Learning- Senior Edition (TOMAL-SE)			
Composite Scores	Standard Score	Percentile	
Verbal Memory	82	12	
Nonverbal Memory	116	86	
Composite Memory	97	42	
Delayed Recall	123	94	
Learning Index	83	13	

Appendix of scores from the TOMAL-SE in March, 2013

WHOLE-WORD TRAINING OF HIGH-FREQUENCY WORDS

Subtests	Scaled Score	Percentile
Facial Memory	9	37
Memory for Stories	7	16
Word List Learning	7	16
Visual Sequential Memory	6	9
Object Recall	6	9
Memory for Location	11	63
Object Memory Delayed	17	99
Facial Memory Delayed	17	99
Memory for Stories Delayed	3	1

In addition to possible impairments in verbal memory and learning, EF is a more severe case of pure alexia than the majority of cases reported in the literature. While similar studies of whole-word training have yielded positive results (Friedman & Lott, 2000: Friedman & Lott, 1997), the same approach may not be appropriate for patients with more severe forms of dyslexia.

Future Research

In looking back on this study, there are a number of aspects that could be improved upon if the study were repeated. One important step that could have benefited this study in many ways is stimulability testing. Informal, dynamic testing of EF"s ability to read rapidly presented words, could have been done before the treatment study began. This may have resulted in a treatment plan more suited to the participant's individual abilities. For example, pre-treatment testing could have been done to determine the optimal duration that the word would be presented. In addition, a simplification in the dependent measures for this study may have resulted in a more representative performance by the participant. During pre- and post-treatment evaluation sessions, EF displayed signs of fatigue and frustration that may have affected his performance. Some of the stimuli in those evaluations were not vital to answering the treatment questions, and could have been eliminated. For example, the control stimuli, List C, as well as the second story reading could have been omitted without significantly affecting the study design.

For future research in this field, one important lesson that can be drawn for this research is the importance of considering verbal memory skills when comparing treatment options for a person with pure alexia. While it is unclear exactly why whole-word training did not yield any gains with the participant EF, poor verbal memory skills may have negatively influenced the outcome of treatment. The implications of this finding should tip off future researchers that whole-word training for those whose verbal memory is impaired.

The most effective approach in treating pure alexia (i.e. letter-by-letter training, wholeword training, or a combination) remains unclear. Much more research is needed in the treatment of pure alexia –especially moderate to severe pure alexia– before an answer to that question will be begin to develop. While the whole-word training approach presented in this study was not successful with EF, this does not mean that similar treatments would not be successful with other patients with severe pure alexia. However, the results do suggest that efforts in researching pure alexia treatment for severe patients may be better directed at improving letter-by-letter reading skills.

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APPENDIX A

The 21 most frequent words in written English, the number of instances in a corpus of 187,272,303 words, and the approximate percentage of written words in American newspapers and magazines.

Word	Instances	Percent
the	10053919	5.4%
and	4785893	2.6%
of	4654561	2.5%
to	4631269	2.5%
а	4587938	2.4%
in	3482180	1.9%
that	1963779	1.0%
for	1735579	0.9%
is	1673946	0.9%
it	1513334	0.8%
with	1314359	0.7%
on	1271008	0.7%
Ι	1213119	0.6%
he	1143222	0.6%
was	1084054	0.6%
as	1080745	0.6%
at	995658	0.5%
are	880170	0.5%
you	867608	0.5%
but	812601	0.4%
from	794002	0.4%

APPENDIX B

List A, B and C. Both List A and List B have five 2-letter words, three to four 3-letter words, and one or two 4-letter words. List A has an average frequency of 12,094 per million and makes up about 12% of written English. List B has an average frequency of 12,215 per million and makes up about 12% of written English. Each word in List C is orthographically similar to a word in one of the target word lists.

LIST A	LIST B	LIST C
to	in	no
of	is	him
it	he	tub
on	as	she
at	by	far
and	the	dad
for	was	era
you	but	saw
are	from	hats
with	that	frog