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Rehabilitation of a case of pure alexia: Exploiting residual abilities

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Abstract

We present a case study of a 43-year-old woman with chronic and stable pure alexia. Using a multiple baseline design we report the results of two different interventions to improve reading. First, a *restitutive* treatment approach using an implicit semantic access strategy was attempted. This approach was designed to exploit privileged access to lexical–semantic representations and met with little success. Treatment was then switched to a *substitutive* treatment strategy, which involved using the patient's finger to pretend to copy the letters in words and sentences. Reading using this *motor cross-cuing* strategy was 100% accurate and doubled in speed after 4 weeks of intervention. We propose that this patient's inability to benefit from the implicit semantic access treatment approach may be in part related to her inability to suppress the segmental letter identification process of word recognition. (*JINS*, 1998, *4*, 636–647.)

Keywords: Pure alexia, Alexia rehabilitation, Letter-by-letter reading

INTRODUCTION

Pure alexia typically refers to a selective reading impairment in which reading is performed by the laborious method of first naming (overtly or silently) the letters that make up each word before the word is recognized. This deficit occurs in the context of relatively preserved writing. The lesion believed to be responsible for generating pure alexia as initially described by Dejerine (1892) and later supported by others (De Renzi et al., 1987; Geschwind, 1965) involves the dominant occipital lobe, usually in the lingual and fusiform gyri and paraventricular white matter of the left occipital lobe (Black & Behrmann, 1994; Damasio & Damasio, 1983). While certain symptoms are shared among cases of pure alexia such as relatively preserved writing and language function as well as increased reading time with increases in word length, other dimensions of the deficit are quite variable. Patients with pure alexia have differed in the areas of implicit word appreciation (Caplan & Hedley-White, 1974; Coslett & Saffran, 1989; Landis et al., 1980; Shallice & Saffran, 1986), reading speed (Bub et al., 1989; Patterson & Kay, 1982; Warrington & Shallice, 1980), word superiority effects (Bub et al., 1989; Reuter-Lorenz & Brunn, 1990), simultanagnosia (Kinsbourne & Warrington, 1962; Warrington & Rabin, 1971; Warrington & Shallice, 1980), and letter naming (Behrmann & McLeod, 1995; Lott et al., 1994; Warrington & Shallice, 1980). This has led some to propose that the underlying mechanisms of these cases (i.e., the particular module of the reading process that causes the difficulty) may differ and thus lead to behavioral differences (Kay & Hanley, 1991; Rothi et al., 1998). Others suggest that the differences across cases result from variations in performance within a single module of the reading process (Arguin & Bub, 1994; Farah & Wallace, 1991). Still others suggest these differences may be due to differences in the selection of strategies or task demands (Coslett et al., 1993; Price & Humphreys, 1992; Speedie et al., 1982).

Differences in performance across reported cases of pure alexia have also led to a number of cognitive neuropsychological explanations for the deficit. The three main hypotheses regarding the nature of the deficit in pure alexia (see Arguin & Bub, 1994; Farah & Wallace, 1991, for more com-

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plete descriptions) are (1) a deficit in low-level visual processes responsible for generating the internal structural representation (Rapp & Caramazza, 1991), (2) a visual perceptual deficit of pattern identification especially for rapidly presented multicomponent input (Arguin & Bub, 1994; Farah & Wallace, 1991; Friedman & Alexander, 1984; Kinsbourne & Warrington, 1962; Reuter-Lorenz & Brunn, 1990) and (3) a deficit particular to the word-form system that prevents access to or activation of word-level representations (Kay & Hanley, 1991; Patterson & Kay, 1982; Warrington & Shallice, 1980). These various hypotheses implicate different aspects of what typical cognitive neuropsychological models of reading refer to as the *visual analysis system* (see Figure 1).

Current theory in cognitive neuropsychology suggests that there are at least two visual analysis mechanisms available to the skilled reader to access stored word representations: (1) a letter-by-letter strategy by which each letter is identified and the composite of these individual letters is assembled to match the stored representation of the word percept, by nature a slow and laborious process; or (2) a whole-word strategy that provides rapid, privileged access to some limited information about the stored word forms based on the parallel mapping of the letter identities as indicated by the visual shape of the word (Bub et al., 1985; Howard & Franklin, 1987; Patterson & Kay, 1982).

According to Howard (1987), words are recognized by the increasingly abstract product analyses of letter features, letters, and component letters by the visual analysis system. Earlier stages of visual processing are feature dependent and therefore affected by alterations of font and case. Later stages are more abstract, yielding what Coltheart (1981) refers to as "abstract letter identifiers," which are type and font independent. The visual analysis system also denotes the position of each letter in the word, tagging it to the abstract letter identities so that words of the same letters but different orders (e.g., tab *vs.* bat) can be distinguished (Ellis, 1993; Hillis & Caramazza, 1992). This abstract level of representation allows for activation of the word recognition units in the orthographic input lexicon regardless of the superficial form of the word. The operations of the visual analysis system are required for both previously experienced words and novel words or pseudo-words. If the word is familiar, the output of the visual analysis system will in turn activate the stored representation. If the pattern from the visual analysis system has not been previously experienced, as in the case of pseudo-words, it can still be processed using a segmented letter-by-letter procedure. Individuals with pure alexia have been noted to rely on a letter-by-letter strategy.

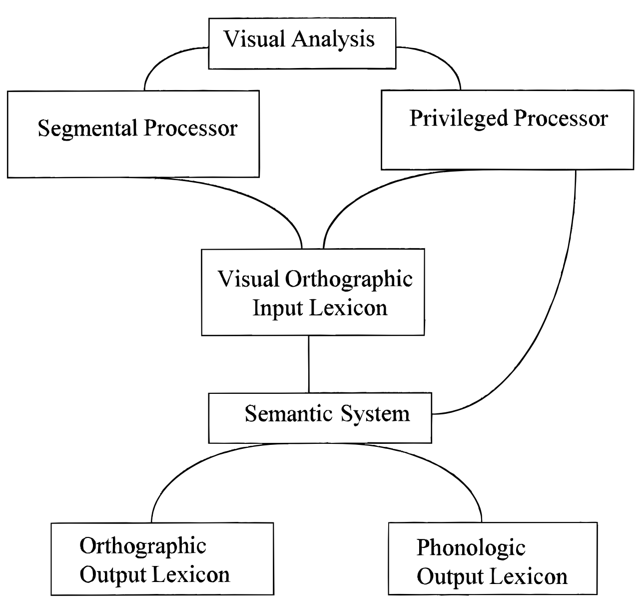

Fig. 1. Simplified model of word recognition and reading aloud.

Recently it has been determined that individuals with pure alexia may retain partial whole-word reading ability, but only at an implicit (subconscious) level (Coslett & Saffran, 1989, 1994; Coslett et al., 1993; Gonzalez Rothi & Moss, 1992). Gonzalez Rothi and Moss suggest that individuals with a letter-by-letter reading form of pure alexia may be trained to disengage the letter-by-letter strategy and recover the whole word strategy, thereby allowing implicit access to meaning from the word form. Other investigators have reported less success using this rehabilitative approach, suggesting that there are subtypes of patients who may not recover implicit access to meaning as the result of utilizing the whole word approach (Rothi et al., 1998). At present there is no way of predicting which individuals with pure alexia may benefit in terms of regaining implicit semantic access when utilizing the whole word approach, and which may require an alternative, substitutive strategy. While the use of cognitive neuropsychological models can be very helpful in identifying which components of a complex process may be deficient, they do not dictate which approaches to rehabilitation may be most efficacious (Behrmann & Byng, 1992; Hillis, 1993; Hillis & Caramazza, 1992). The clinician still must determine if rehabilitation should target the underlying deficit or exploit residual abilities (Behrmann & McLeod, 1995).

Rothi (1992, 1995) maintains that the physiologic state of the recovering system should be considered when devising a rehabilitation program. Specifically, she suggests that restitutive treatments (treatments designed to restore the impaired ability using the same functional processes in the same manner, as utilized premorbidly) are strategic approaches to treatment that reflect an assumption that functional recovery will occur based upon reconstructive processes within the nervous system (e.g., reactive synaptogenesis, dendritic sprouting). These treatment strategies would be better applied during the early stages (postonset) of recovery, when the *greatest* amount of physiologic restoration is most likely to occur. Alternatively, substitutive treatments may be effective without temporal limitations but are not likely to be as efficient or as elaborate as the original system (for example, gestural systems or speech synthesizer computers *vs.* human speech). With respect to substitutive therapies, they may be *vicariative* or *compensative* in nature. Vicariative

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treatments recruit other systems or processes not premorbidly utilized in support of the target behavior which are intended to permanently support the reorganized system or process (Rothi, 1992, 1995). With respect to the reading system, individuals with pure alexia who use a letter-by-letter reading strategy are using a substitutive strategy, in that the naming of letters is a means of compensating for failure to activate the conventional method for decoding written input. However, many individuals with pure alexia find this strategy too slow and complicated by memory impairments; thus, it is not a functional substitutive approach. Reliance on implicit semantic activation may be more of a restitutive strategy in that this type of activation is believed to be part of the normal process for word recognition (though not at the conscious level) and may be an obligatory aspect of skilled reading. However, reliance on this component of the system may only be necessary when explicit word identification is demanded (Coslett et al., 1993). Thus the application of whole word training to remediate pure alexia, because it is a restitutive strategy, may be more time-limited than other rehabilitation strategies that are more substitutive in nature.

We present the results of two approaches to remediation of an individual with pure alexia that was $2\frac{1}{2}$ years post-onset. We initially attempted to exploit implicit, whole word semantic activation. The use of multiple baseline single-subject design allowed us to determine relatively rapidly that this approach was not effective. We then attempted a more substitutive approach to treatment, exploiting residual abilities identified in the evaluation of this individual's reading impairment. Our rehabilitation efforts attempted to model the clinical application of current reading research paradigms and theory, modified to suit the clinical setting.

METHODS

Patient

V.T. is a 43-year-old, right-handed woman with a history of an ischemic stroke in August 1994, which resulted in a dense right homonymous hemianopsia, alexia, "confusion" difficulty with speech articulation, difficulty with balance, and personal memory loss. She apparently suffered a second CVA in December 1994, which reportedly did not result in any additional symptoms. MRI scans (Figure 2) after the first event revealed a large left-hemisphere occipital stroke. There was no change on MRI following the second event. The le-

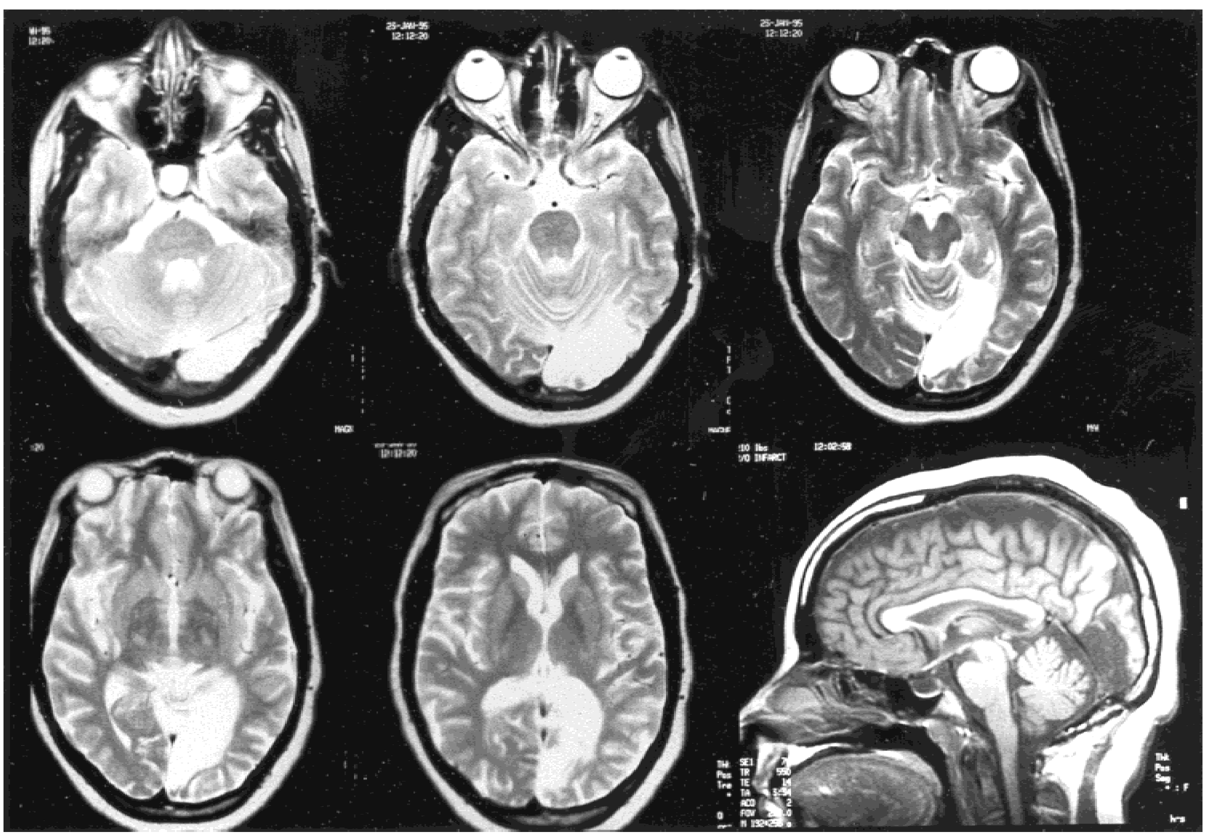

Fig. 2. T2-weighted axial MR images and T1-weighted sagittal MR image of the brain for Patient V.T. Right side of the brain is seen on the left or axial images—sagittal image is of the left hemisphere. Infarction is seen in the territory of the left medial branch of the posterior cerebral artery (calcarine and parieto–occipital arteries). Infarction includes parts of Brodmann's areas 28, 31, and 18, and the left cerebellar hemisphere, as shown. Cortical areas of infarction include the lingual gyrus and parts of the fusiform gyrus and cuneus, and the retrosplenial area.

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sion was plotted onto matched images (Damasio, 1995) and appeared to be in the territory of the left medial branch of the posterior cerebral artery (calcarine and parieto–occipital arteries), affecting Brodmann's areas 28, 31, and 18, and the left cerebellar hemisphere. Signal was consistent with infarction and cortical areas of infarction included the lingual gyrus and parts of the fusiform gyrus and cuneus, and the retrosplenial area.

V.T. stated that she holds an undergraduate degree in organic chemistry and that prior to her strokes she was employed as a synthetic organic chemist and lab supervisor. She was fluent in English (as her primary language), Spanish, and Italian. V.T. received no speech–language therapy immediately after her strokes, but did attend a rehabilitation center for the visually impaired, apparently because of a visual field cut and because of difficulty she had "making sense" of the environment. There she was trained in mobility and daily living skills. She reported having been trained to scan each visual quadrant to "piece together the landscape." She completed 1 year of training in reading Braille. At the time of this investigation, she was able to read Braille at Grade 2, at 46 words/min. In October of 1995, at 14 months postonset, she was referred to the Georgia State University Speech–Language-Hearing Clinic for a speech and language evaluation due to concerns about a persistent speech difficulty. This evaluation revealed mild consonant imprecision, particularly for lingual phonemes and consistent with tongue weakness. She also presented with decreased respiratory support for speech characterized by decreased loudness in connected speech. There was no evidence of aphasia. Her score on the Boston Naming Test (BNT; Kaplan et al., 1983) was 59 correct out of 60 possible, and on the Western Aphasia Battery (WAB; Kertesz, 1982) she received a score of 9.7 out of 10 possible, consistent with normal performance. Her repetition was intact, written expression was flawless, and there was no evidence of syntax impairment in either spoken or written expression. In contrast, reading was found to be severely impaired. V.T. reported that she was completely unable to read. On the Boston Diagnostic Aphasia Examination (BDAE; Goodglass & Kaplan, 1983) she was unable to match printed words to pictures, point to named printed words, or read single words aloud. She was unable to read her written description of the "cookie theft" picture from the BDAE that she had written minutes earlier. While able to select an exact match of printed words from a visual array of five words, she was unable to name the words and she was also unable to match letters or words that differed in fonts or case. When asked to trace the words with her finger, she was able to recognize each letter and read the word aloud. She was quite surprised at this ability to recognize words once she traced them, stating that she "hadn't thought of trying that." She reported that she had tried to relearn to read by watching children's educational TV and had attempted commercially available, educational programs for reading, but all were unsuccessful.

Following this evaluation, V.T. received speech therapy to improve her mild unilateral upper motor neuron dysarthria (Duffy, 1995). Her alexia was not treated however, as she indicated that she did not want to address her reading difficulty at that time. Following discharge from speech therapy, she was then referred to the current investigators for a complete evaluation of her reading and possible remediation of her alexia. While V.T. had achieved fair proficiency in reading Braille, she was limited by the lack of available Braille materials for everyday activities (dining in restaurants, shopping, sheet music, etc.), and expressed a desire to try to regain reading ability for print.

Assessment

In an effort to determine which aspect or aspects of the reading system were impaired, V.T. underwent a number of formal and informal measures. These will be discussed as they relate to the cognitive model of reading presented in Figure 1.

Visual perceptual processing

Visual field testing revealed a dense right homonymous hemianopsia. V.T. did not respond to stimuli presented in both the right upper and lower quadrants. There was no evidence of hemispatial neglect as measured by line bisection and cancellation (Heilman et al., 1993). Color recognition presented to the left visual field was flawless.

The Visual Object and Space Perception Battery (VOSP; Warrington & James, 1991) and the Motor-Free Visual Perception Test–Vertical (MVPT–V; Mercier et al., 1997) were administered to identify the presence of visual perceptual deficits. All stimuli were placed to the left of midline to accommodate V.T.'s right visual-field deficit. V.T. completed all subtests of both measures within the normal range of performance, suggesting that she did not have difficulty with the perception of shapes, degraded stimuli, unusual views or visual memory for shapes (see Table 1). Visual object recognition and naming were also within normal limits as suggested by a score of 59/60 on the BNT. This test was given under two conditions: once with unlimited presentation time, and on another occasion with stimulus presentation time limited to approximately 500 ms. There was no difference in V.T.'s performance in the two conditions.

Letter recognition was severely impaired and V.T. was not able to match any letters or words that varied in font or case on the symbol and word discrimination subtest of the BDAE. She was able to indicate presence of the letter *X* in letter strings ranging from three to seven letters with 100% accuracy, though response times were slow, and increased when the *X* occurred toward the end of the letter strings. In contrast to letter strings, identification of the presence of a zero or an *X* in a string of numbers was rapid and flawless, and there was no indication of an effect of place for number strings. On number recognition tasks, V.T. was able to read aloud numbers from one to five digits (e.g., 16,789) accurately and without hesitation. The inability to recognize or match letters of varying fonts and case is suggestive that

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Table 1. Visual Object and Space Perception Battery (VOSP) and Motor-Free Perception Test–Vertical Format performance by V.T.

Test–Subtest	% Correct
VOSP	
Incomplete Letters	100
Silhouettes	90
Object Decision	90
Progressive Silhouettes	n/a
Dot Counting	90
Position Discrimination	95
Number Location	100
Cube Analysis	100
MVPT–V	
Exact Figure Matching	100
Altered Figure Matching	100
Degraded Figure Matching	88
Visual Memory	91
Figure Discrimination	100

V.T. has a deficit at the level of the abstract letter identifiers, which normally should be able to recognize letter regardless of variations in font or case. Her ability to match exact items suggests that she was able to perceive the stimuli.

Access to the orthographic input lexicon

V.T. was able to recognize all orally spelled real words (regular and irregular) and nonwords from the Battery of Adult Reading Function (BARF; Rothi et al., 1986) presented to her. In striking contrast, V.T. was unable to read aloud any of these same stimuli when presented in printed form. However, she was able to "read" aloud the same sets of words and nonwords presented visually when allowed to copy the target on her hand or lap, or to visually "trace" the words (which involved movement of her head).

Visual imagery for letters and words

V.T. was able to answer questions about the physical attributes of named letters (e.g., "Indicate if a stated upper case letter contained all straight lines, all curved lines or both") with 100% accuracy. She was also able to indicate which spoken word had more letters (e.g., "*fan vs. phone*") with 100% accuracy, suggesting preservation of visual imagery for letters and words. Some have suggested that preservation of visual imagery suggests that the store for letters and words may be preserved (Crary & Heilman, 1988). However, when we asked V.T. how she completed these imagery tasks, she said she "wrote" the letters or words mentally, thus determining the answer. V.T.'s ability to access the phonologic form of words and nonwords from auditory and motor–kinesthetic input, but not from visual input alone, suggested a deficit in accessing the visual orthographic input lexicon (i.e., getting from the visual stimulus to the orthographic lexical store). Her ability to answer questions based on the form of the letters as in the imagery task can be explained by her self-reported use of what we will call a *motor cross-cuing strategy*. When using this strategy, she may be depending on the motor–kinesthetic sense of writing the letters or words to identify them or answer specific questions about form.

Visual orthographic input lexicon

V.T. was observed to use this motor cross-cuing strategy to complete selected lexical–orthographic subtests on the Psycholinguistic Assessment of Language Processing in Aphasia (PALPA; Kay et al., 1992). Placing her hand in a pen-holding position with her index finger extended and rapidly copying the shape of each letter in each word, she was able to "read" all of the items without difficulty. These data are presented in Table 2. Performance on these tasks suggests that once able to access the orthographic input lexicon, her lexical knowledge was intact.

Semantic system

V.T.'s ability to comprehend what she "read" was tested informally. When answering yes–no questions about material she read that was single sentence length, she was 100% accurate. It would appear that once she was able to identify a word using her motor cross-cuing strategy, she demonstrated no difficulty with comprehension of that word.

Orthographic output lexicon

V.T. demonstrated intact writing ability for single words and paragraphs. She was able to write all of the items on the BARF (Rothi et al., 1986) and her written descriptions of the "cookie theft" picture from the BDAE (Goodglass & Kaplan, 1983) and the "kite" picture from the WAB (Kertesz, 1982) were accurate in spelling, sentence structure, and substance. V.T. reported that she often writes letters to friends, though she is unable to read what she has written.

Phonemic output lexicon and grapheme-to-phoneme conversion

Based on preserved abilities in spoken discourse, naming, picture description, and repetition, V.T. demonstrated no ev-

Table 2. PALPA Performance by V.T. using motor cross-cuing strategy

Subtest	% Correct	Time
Visual Lexical Decision		
Illegal Nonwords	100	11:46
Imageability and Frequency	99	25:15
Morphology	98	13:04
Spelling–Sound Regularity	100	10:18
Homophone Decision	100	14:42

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idence of difficulty at the level of phonologic output. When observed using the motor cross-cuing strategy V.T. was able to read regular and irregular words of varying frequencies and imageability aloud, albeit slowly without error. Using this strategy she was also able to read nonwords aloud, suggesting intact grapheme-to-phoneme conversion once the graphemes were accessed. Implicit lexical access to semantics was assessed by asking V.T. to sort written words based on some semantically based distinction, such as *living versus nonliving*, or *edible versus nonedible*. Each category contained 10 words. The words were printed individually on index cards, and presented one at a time to the left of V.T.'s midline for approximately 1 s. The investigators originally planned to present each stimulus for approximately 500 ms, but V.T. complained that she "could not even see the word" during that short time period; thus, the presentation time was increased to 1 s. Each trial consisted of a block of 20 words (two sets of 10 words each). Results of this testing are presented in Table 3. These results suggest that V.T. was basically at chance performance for most of the trials, indicating a relative lack of implicit access to semantics *via* visual orthographic input.

Reading speed

Measures were taken of the length of time needed for V.T. to read single words and paragraphs aloud. It should be noted that the only way V.T. was able to complete these tasks was by using the motor cross-cuing strategy. For comparison purposes we also obtained measures of her reading speed for Braille, which she did tactually. These data are presented in Table 4. V.T.'s pattern of "reading" performance on these tasks suggested that her reading rate was markedly slowed and negatively affected by word length. A word length effect is consistent with letter-by-letter reading frequently described in pure alexia, where the individual names each letter in the word in order to recognize and name the word. V.T.'s strategy of "copying" the words for recognition also appeared to be a letter-by-letter strategy in that she would copy each letter, and while she would not name each letter aloud, the more letters in a word the longer it took for her to copy it. Her reading speed was not improved by increasing the size of the letters. While her reading of handwritten script seemed to be faster than printed text, this was likely due to the fact that she was reading familiar text that she herself had written a short time earlier, whereas, the printed text she read was novel. Finally, her Braille text reading was not much faster than her reading using the motor cross-cuing strategy. V.T.'s skill level with Braille still required reading each letter symbol individually before she recognized the word; thus Braille reading also appeared to be at a letter-by-letter decoding level.

Table 3. Subthreshold lexical–semantic access performance by V.T.

Trial*	Categories	% Correct**
1	Edible *vs.* nonedible	40
2	Fruit *vs.* vegetable	65
3	Edible *vs.* nonedible	85
4	Edible *vs.* nonedible	60
5	Living *vs.* nonliving	65
6	Edible *vs.* nonedible	45
7	Living *vs.* nonliving	65

*Each trial contained 20 cards per sort.
**Chance performance = 50%.

Table 4. Reading speed performance by V.T.

Reading material	Reading speed
Words	
Two- to three-letter single words	2.6†
Four- to six-letter single words	4.1†
Seven- to nine-letter single words	6.3†
Text	
Printed paragraph 18-point	13 WPM*
Printed paragraph 10-point	11 WPM*
Handwritten picture description	29 WPM*‡
Braille: Grade II	14 WPM

Note. WPM = words/min.
†Average speed recorded in number of seconds per word.
*All items read using motor cross-cuing strategy.
‡V.T. read handwritten paragraph shortly after writing it.

Summary of alexia testing

The results of the initial alexia assessment suggested that V.T. had alexia without agraphia, with preservation of the orthographic lexical–semantic system. We hypothesized that her deficit was somewhere in the visual analysis system, prior to accessing the visual orthographic input lexicon. She was unable to realize the abstract letter identities believed to be the output of the visual analysis system, either because she could not recognize the visual input as letters, or because she could not correctly identify the position of the letter in the word, or both (Ellis, 1993; Hillis & Caramazza, 1992). Evidence of impairment of identification of individual letters is found in her inability to match letters of different case and font. Anecdotally, V.T. complained that she was unable to determine the boundaries of each letter, and the boundaries between words. This suggested difficulty processing the multiple features associated with letters and words, similar to the "alexia simultanagnosia" reported by Kinsbourne and Warrington (1962), again implicating early visual analysis. V.T. differs from many of the reported cases of pure alexia in the literature in that she was not able to name individual letters by visual input alone. She was able to achieve letter and word recognition by utilizing a motor cross-cuing strategy, which revealed a sensitivity to word length consistent with letter-by-letter reading. Results of her poor ability to semantically categorize printed words presented at subthreshold exposure times suggested that the privileged processor was not reliably available.

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Rehabilitation Program

Having been shown how she could access words using the motor cross-cuing strategy, V.T. was anxious to begin a program for reading rehabilitation. There were three phases to her rehabilitation program. Each of these phases are described separately and the data are presented in Figure 3.

Untreated sentence probes

In order to assess the effect of treatment on untreated exemplars, data were collected on V.T.'s speed of reading sentences aloud that were not part of the corpus used in treatment. Reading speed data were collected prior to the initiation of the first phase of training, and at the beginning of each therapy session. The purpose of the probes was to ensure that these behaviors were stable and in turn, determine if any changes observed during the therapy interval generalized to untreated sentences. Ten 10-word sentences were randomly chosen from the first 60 10-word sentences on the Assessment of Intelligibility of Dysarthric Speech (AIDS; Yorkston & Beukelman, 1981). A different set of sentences was used each day to prevent the influence of familiarity on reading speed. V.T.'s reading of each day's set was tape recorded for scoring at a later time. Reading speed was timed for each sentence using a stopwatch from the moment the sentence number was named by the examiner until V.T. completed reading the sentence aloud. These speeds were then added together to reach a total time for reading aloud per 100 printed words. The reading rate was then converted to words per minute for ease of interpretation (see Figure 3). V.T. used the motor cross-cuing strategy to read the sentences, as she was unable to do the task otherwise.

As a control measure to assure that any changes in V.T.'s performance were not simply due to the effects of practicing the tasks regularly, V.T. was timed reading a Braille passage aloud every other session. It was predicted that if treatment was effective, its effect would be specific to the visual reading of sentences and not to Braille reading. The Braille readings were on a fifth-grade reading level. The time in seconds per 100 words of Braille read aloud was recorded with a stopwatch for comparison to the reading measures. This was also converted to words per minute to make interpretation easier.

Phase 1: implicit semantic access training

The purpose of this phase of treatment was to train V.T. to regain implicit lexical semantic access by using the restitutive strategy of a word form or whole word visual analysis (Gonzalez Rothi & Moss, 1992; Rothi et al., 1998). In order to prevent or disengage the explicit, letter-by-letter analysis, written words were visually presented at a subrecognition threshold, which for V.T. was determined to be less than 1 s. V.T. was asked to report a semantic category distinction for the target word based on this exposure (e.g., the word presented was *apple*, and V.T. was asked to determine if the word was edible or nonedible). Category groups with ten

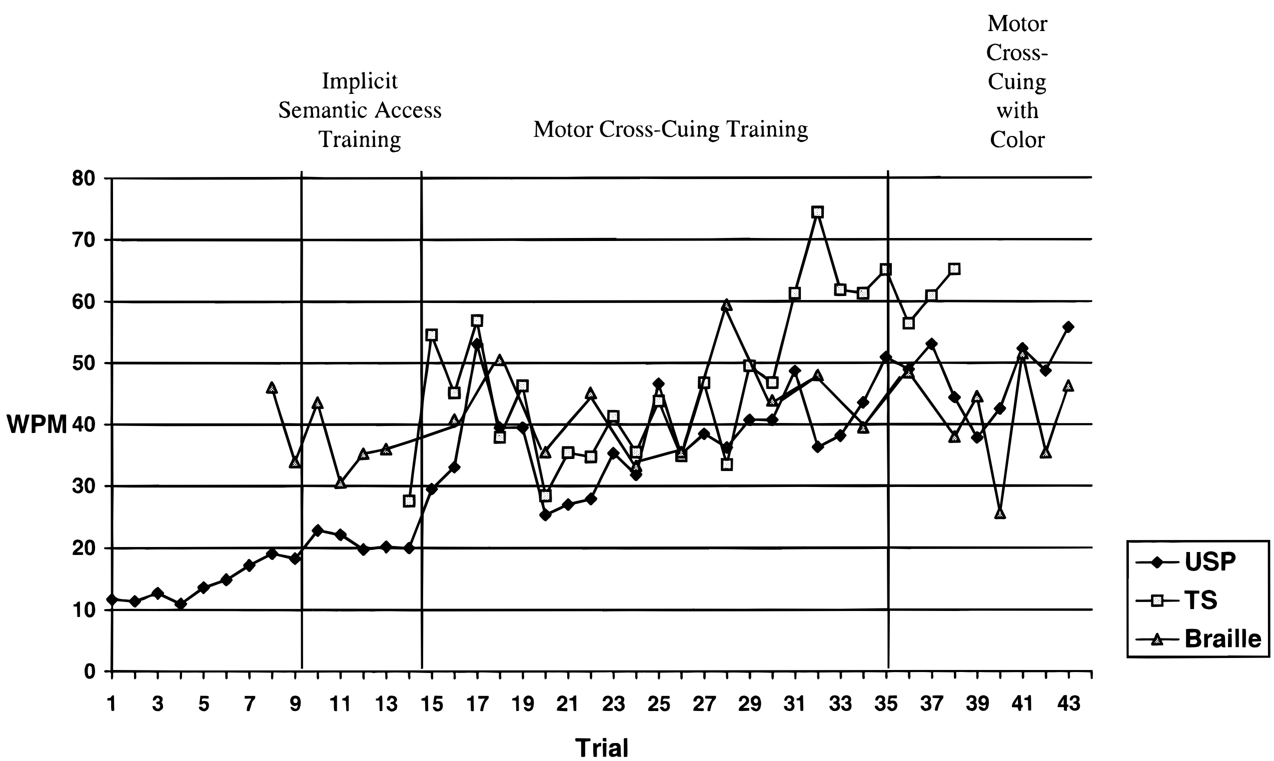

Fig. 3. Reading speed of untreated sentence probes (USP), treated sentences (TS) and Braille in words per minute (WPM) across all treatment phases.

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words each were chosen, and the words were printed individually in lowercase letters on 7.6 × 12.7 cm index cards. Two random categories were chosen and mixed together. The cards were shown one at a time to V.T. for approximately 1 s each and then removed. Prior to the initiation of treatment, V.T.'s accuracy on this task had been determined as part of the alexia assessment (over three consecutive sessions) and ranged from 40 to 85% (see Table 3). With the exception of one trial at 85% accuracy, V.T.'s semantic accuracy on this task prior to treatment was basically consistent with chance performance. Despite this less than promising performance, we attempted a trial period of treatment as it was not clear from previously reported cases if evidence of implicit semantic access was needed for training using semantic categorization to be effective. Four different semantic categories were selected for treatment. Words from two sets of categories were shown for less than 1 s of exposure time, and V.T. was asked to force her eyes to scan the whole word presented to her and to take note of the word's shape. After the word was removed, V.T. was asked to indicate to which category the word belonged. She frequently complained that she had not seen the word long enough to tell, and she was encouraged to make her best guess. Treatment involved the clinician providing verbal feedback to V.T. regarding the accuracy of her response. Each treatment trial consisted of 20 items, and between two and five trials were completed per therapy session. For approximately half of the trials, V.T. was given immediate verbal feedback for each item by the clinician. For the remaining trials, the clinician gave verbal feedback only after a block was completed. Regardless of the type of feedback she received, V.T.'s performance on the treatment sets ranged from 65% accuracy to 30% accuracy. V.T.'s performance on the pretreatment sets also remained at chance. V.T. expressed feelings of frustration due to her lack of success during these sessions. Despite being reminded to look at the whole word, and despite being presented the stimuli for short durations that prevented visual segmentation, she continued to identify the initial letters. She repeatedly stated that her performance would improve if she could use her finger to read. Due to the lack of favorable results and V.T.'s dissatisfaction with the program, the training was abandoned. Visual inspection of V.T.'s reading speed for the untreated sentence probes (Figure 3) during this phase of treatment suggests that implicit semantic access training did not have an impact on V.T.'s reading speed for untrained stimuli.

Phase 2: motor cross-cuing training

V.T. expressed a desire to incorporate the motor cross-cuing technique into her reading attempts and therefore we focused on this method next. Therapy sessions involved V.T. using her finger to trace the motor output for word recognition. This substitutive technique is similar to that reported by Lott et al. (1994) except that their patient "copied" the letters on the palm of the hand, yielding both tactile and motor feedback. While V.T. used this strategy to complete the baseline and control measures, she had not begun to use it functionally. She was trained to trace the shape of each letter on a hard surface (e.g., the table or a book) with her right index finger until she had traced all letters in the word. V.T. did not require tactile feedback to recognize the word, nor did she name the letter (either aloud or silently). She appeared to be able to recognize the letters and words as a result of the motor activity (not unlike the use of pseudodialing to recall a touch tone phone number). Training materials consisted of 100 five-word sentences and 100 six-word sentences taken from Yorkston and Beukelman (1981). V.T. was asked to use the motor cross-cuing strategy to read the sentences aloud and to do so as quickly as she could. Training was performed with blocks of 10 sentences, and her reading speed was recorded for each sentence in the same manner as used in the baseline condition. An average speed was taken for each block, and treatment consisted of the clinician providing V.T. with feedback regarding her reading speed and encouraging V.T. to rapidly "push through" the words in the sentence. Treatment was conducted in 1-hr sessions, four times a week for $4\frac{1}{2}$ weeks. After 4 weeks of treatment it appeared from visual inspection of the data that V.T. was no longer making further gains in reading speed for treated or untreated sentences. We suspected that V.T. had possibly achieved maximum gains using this substitutive strategy, and we initiated the next phase of treatment at that time.

Phase 3: color spacing plus motor cross-cuing

As V.T. progressed through the motor cross-cuing treatment, she continued to report difficulty separating words one from another (i.e., identifying where one word ended and the next word began). At the conclusion of Treatment Phase 2, we began the third phase of therapy. New sentences, these seven and eight words in length, were selected from Yorkston and Beukelman (1981) for Phase 3 training. The treatment in Phase 3 was similar to Phase 2 in that V.T. continued to use the motor cross-cuing strategy, was encouraged to read the sentences as quickly as she could, and was given feedback as to her reading speed, but we added color highlighting in the space between each word in an effort to facilitate identifying the boundaries between words. Using seven- and eight-word sentences, the spaces between words were colored using pastel markers. V.T. continued using the motor cross-cuing strategy in this phase, which she insisted was necessary for her to be able to identify the words. This was done for six sessions in the clinic over a 2-week period of time. Visual inspection of those data both for untreated and treated sentences suggested no improvement in reading speed after six sessions, and treatment was discontinued.

RESULTS

Response to the three phases of the rehabilitation program was measured by monitoring V.T.'s reading speed for the

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untreated sentence probes. Visual inspection of the data (see Figure 3) suggests a modest rise in performance with simple repetition of the untreated sentence probe measure during the initial no treatment period; however this leveled off during the first treatment phase and remained relatively stable prior to the initiation of the second phase.

Phase 1: implicit semantic access training

In this phase of therapy, by reducing stimulus exposure duration thereby limiting segmentation and requiring a semantic decision, we hoped to exploit implicit semantic access, and at the same time increase reading rate and proficiency. As stated above, this phase of rehabilitation had little demonstrable effect on reading speed. Training lasted for six sessions, but was quickly abandoned as V.T.'s performance remained at chance accuracy in choosing the correct semantic category and she expressed tremendous frustration over her performance. She frequently complained that she was unable to see the letters, and stated that without using her finger she was not able to know anything about the meaning of the words.

Phase 2: motor cross-cuing

In contrast to the implicit semantic access training, there was a marked improvement in V.T.'s reading speed soon after the initiation of the next treatment, motor cross-cuing. Gains in reading speed using this strategy continued and remained stable throughout the remainder of the study. V.T.'s average reading speed prior to the initiation of cross-cuing training was 19.96 words/min. Mean reading speed for the last eight sessions of Phase 2 was 44.55 words/min, more than double the pretreatment speed. Because we were concerned that improvements in V.T.'s reading speed may have been influenced by repeated exposure to the untreated sentence probes even though they were randomly selected each session, reading speed on an additional 40 10-word sentences having the same characteristics as the untreated sentence probes (Yorkston & Beukelman, 1981) was sampled during the last week of Phase 2. Mean reading speed for this set of sentences was 43.58 words/min, which was comparable to the untreated sentence probes, suggesting that V.T.'s improvement could not be simply attributed to repeated exposure to the probe sentences.

Phase 3: color spacing plus motor cross-cuing

Reading with color spacing between words appeared to have little additional effect on reading speed. Average reading speed with color spacing was 49.02 words/min, relatively consistent with V.T.'s performance at the end of Phase 2.

In summary, these data suggest that reading speed for V.T. was not affected by the implicit semantic access training or the addition of color spacing to emphasize word boundaries, but that practice in the use of motor cross-cuing resulted in a substantial increase in reading speed. These gains cannot be explained simply on the basis of repeated practice because reading speed was stable on the untreated sentences prior to the initiation of treatment and a marked improvement in reading speed on those same sentences was not demonstrated until formal treatment was initiated. Neither can they simply be attributed to generalized practice effects, because reading speed for Braille remained constant throughout the study, averaging 46.0 words/min at the beginning of the study, and 44.4 words/min at the study's completion. Furthermore, these gains appear to have generalized beyond the sentences used as treatment probes, since performance on a novel set of sentences having similar characteristics also showed the same relative gains in reading speed. However, it is also clear from the data that gains in reading speed were limited by this technique. Visual inspection of the data suggests that improvement in reading speed soon stabilized after the initiation of training, and did not improve further with continued practice. This is not surprising given the fact that V.T. was unable to compress her use of the motor cross-cuing strategy to where she did not have to pseudo-write each letter. This strategy is by nature a letter-by-letter strategy, but instead of naming the letters, V.T. wrote each of the letters and only in doing so was she able to achieve word recognition. It may be that she was reading as fast as this strategy would allow. At the beginning of the study V.T.'s single-word reading performance suggested a letter-by-letter pattern in that reading speed increased for longer words (see Table 4).

While reading speed at the termination of treatment was still much slower than normal performance, V.T. was very pleased with her progress and had begun to read again for pleasure. She reported that she was now able to read whatever she wanted as long as she could take her time. She indicated a preference for reading using the motor cross-cuing strategy rather than Braille in that it was readily available to her at any time.

DISCUSSION

We have presented a case of pure alexia who at greater than 2 years postonset had failed to relearn reading independently. This case is presented to illustrate the clinical application of current reading theory on rehabilitation of pure alexia. We attempted to use our understanding of a normal reading model to drive the focus of V.T.'s rehabilitation. However, our effort to exploit implicit activation of lexical–semantic representations by reducing stimulus exposure durations thereby disengaging visual segmentation, and requiring a semantic decision, was not successful. In contrast, after a fairly short course of treatment V.T. was able to utilize a substitutive strategy of pretending to copy the written word and thus cross-cue and activate stored lexical representations for word recognition. This strategy is not one suggested by theoretical models of reading, but rather one suggested by exploring the individual's performance and exploiting residual abilities to substitute for impaired ones. This is not to suggest that theoretical models are not useful in the rehabilitation of pure alexia or any other cognitive impair-

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ment, but only that they do not ultimately dictate which procedures or strategies will be efficacious for the individual. The reading model was useful in guiding a thorough assessment of V.T.'s reading system, and in so doing, we were able to identify potential approaches to rehabilitation. By using a controlled, single-subject design, we were able to determine relatively quickly which approaches were efficacious, and which were not. Unfortunately, from a theoretical standpoint, more questions are raised by this case than answered.

The first question is why was the implicit semantic access treatment unsuccessful? One possibility is that this automatic process was lost as a result of the brain damage sustained by V.T., a possibility that is supported by V.T.'s poor semantic category performance prior to the initiation of implicit semantic access treatment. Ellis et al. (1988) suggested that the visual perceptual processors of the left and right occipital lobes differ in how they function. They proposed that the left occipital region is a rapid, parallel visual processor of letters and words that has a privileged access to the word recognition system. They also suggested that both hemispheres have a second, "segmental" processor that processes letter strings in an "ends-in" fashion. In the case of a left occipital lesion, the right hemisphere's segmental processor can still operate, but the privileged parallel processor in the left hemisphere cannot function. It may be that V.T. no longer had access to any function of the left hemisphere privileged processor, as her lesion was quite extensive.

While this explanation neatly accounts for much of V.T.'s difficulty, it cannot explain why she was not able to recognize letters or words unless she was allowed to "copy" them first. This difficulty suggests that she was unable to access any stored orthographic knowledge from the visual input. She seemed to have lost the ability to extract the features of letters necessary to activate the representation, or her recognition was so slowed that the abstract percept was lost. This was not the case for all visual input, since she had no difficulty recognizing line drawings or numbers. Her difficulty seems to be most consistent with the alexia simultanagnosia described by Kinsbourne and Warrington (1962). How then did the strategy of "copying" the words assist in V.T.'s ability to read? V.T. demonstrated flawless recognition of words spelled to her, indicating that she could activate those stored representations from other input modalities. It would appear that the motor activity of copying the letters yielded an alternate modality of activation of the lexicon. V.T. was then able to exploit this ability to access words at a rate of approximately 45 words/min. Others have reported similar "copying" strategies (see Lott et al., 1994 for one example) but their programs always tried to fade the use of the cuing strategy to depend on visual input. V.T. was no more able to recognize visual letters at the end of the study period than she was at the beginning. It remains to be seen if there is some way to treat the impaired visual analysis system to allow for sufficient activation for word recognition.

Another question raised by this case is why did the implicit semantic access training fail for V.T., having been successful in other cases of pure alexia rehabilitation. Rothi et al. (1998) suggest that their treatment failure provides evidence that not all cases of pure alexia are the same and thus not all will respond to the same treatment. This is supported by the variable results in treating pure alexia using other types of intervention, such as multiple oral rereading (Beeson, 1998; Moody, 1988; Moyer, 1979; Tuomainen & Laine, 1991). We would agree with this position, though it does not necessarily indicate that the nature of the deficit is different simply because the response to treatment varies. Coslett et al. (1993) have demonstrated that even within the same patient, the strategy used to activate the word recognition system can vary. They suggest that letter-by-letter reading and whole word reading are two distinct and incompatible processes. They were able to induce their patient to switch between the two strategies by altering the task demands. When they required the patient to name the word aloud, the patient reported being aware of switching to a letter-by-letter strategy. Conversely, when the task required a quick lexical or semantic decision, the patient adopted a rapid, whole word strategy. We would suggest that not all patients are so readily able to switch between the two routes and that the failure of some letter-by-letter readers (and in this case a letter-to-motor reader) to respond to whole word intervention is an inability or refusal to suppress the letter-by-letter strategy. In the case of V.T., this may have been secondary to the severity of her visual analysis impairment, which may have prevented even implicit access to the orthographic input lexicon. Conversely, V.T. may have been reluctant to suppress a strategy that had validity to her (i.e., motor cross-cuing strategy, which had already been demonstrated to have the potential to meet her functional reading needs) for an approach that seemed relatively impossible (i.e., the rapid processing of words that she could not even perceive). Finally, we cannot rule out the possibility that improvement in implicit, semantic, access may be more time locked, due to the restitutive nature of the approach, and that V.T.'s lack of success using this technique was influenced by the chronic nature of her deficit.

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