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The Evolution of Pure Alexia: A Longitudinal Study of Recovery

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This case report documents the partial recovery, over a 12-month period, of pure alexia in an adult female following a left occipital infarction. Measures of speed and accuracy were obtained on an oral reading and a lexical decision task immediately postonset and then on 10 subsequent occasions. Explicit letter-by-letter reading was observed only during the first week poststroke but a significant effect of word length was seen in all testing sessions. Reading accuracy was relatively good at all stages and reading latency showed a remarkable decrease over time but did not reach normal reading rates. The inability to use higher-order orthographic knowledge, as manifest in the absence of a word superiority effect, was still noted at one year postonset. We therefore concluded that the change in behavior was attributable to increased proficiency in the use of the adaptive letter-by-letter procedure rather than to the resolution of the underlying

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deficit. It is suggested that longitudinal neurobehavioral studies add to our understanding of the alexic deficit and provide insight into the recovery process.

Systematic longitudinal studies of recovery from alexia are sparse, unlike the many detailed reports of recovery from aphasia. The early anecdotal accounts of alexia recovery are brief and provide contradictory accounts, with some patients demonstrating minimal or no recovery and others improving with specialized training (Benton, 1964). More recently, Marshall and Newcombe and their colleagues (Marshall, Newcombe, & Hiorns, 1975; Newcombe, Hiorns, & Marshall, 1976; Newcombe & Marshall, 1973; Newcombe, Marshall, Carrivick, & Hiorns, 1976) presented a series of investigations in which they plotted recovery curves in several patients with acquired dyslexia. Both qualitative and quantitative differences emerged over time, with changes in the shape of the recovery curve and in the nature of the error patterns. A fundamental point made by these authors is that in-depth longitudinal studies provide a crucial source of evidence which increases our understanding of physiological recovery mechanisms. Knowledge conerning the potential for and constraints on recovery is critical for the purpose of prognosis and remedial intervention. In this paper, we track the recovery of a single subject with acquired letter-by-letter dyslexia in order to document subtle changes in reading performance over time. In addition to providing longitudinal empirical data, we also attempted to examine whether the changes in performance were attributable to the resolution of the underlying functional lesion which gave rise to the reading deficit or whether the observed changes resulted from increased efficiency in the use of the adaptive letter-by-letter reading procedure.

Recent research on spontaneous remission in acquired neurobehavioral disorders has generally included reading alongside other language functions. These studies have documented the reading deficit in relation to Broca's and Wernicke's aphasia and have tended to focus on the overall pattern of performance in relation to etiology, lesion size, and other possible prognostic factors (Kertesz, 1979, 1984). Since reading shares some, but not all, cognitive processes with other language functions, the recovery of reading, in many instances, is poorly aligned with the recovery of these other functions (Gardner, Denes, & Zurif, 1975). The study of the recovery of reading therefore merits independent consideration. Pure alexia, in which the reading disorder is largely unaccompanied by other cognitive deficits, provides an important opportunity for such study. For example, Newcombe et al. (1974, 1976) systematically documented the behavior of MB, a patient with residual dyslexia over a period of 160 weeks, following evacuation of a large left occipital intracranial abscess. At 3 months postsurgery, MB was able to name letters but not to read words, a pattern typical of alexia without agraphia. Over time, the number of MB's error responses decreased, reaching an asymptote around 60 weeks postsurgery. Interestingly, the type of error observed changed across testing periods, somewhat resembling the pattern of initial reading acquisition in children. Initially, MB attempted to spell the words out loud and failures of visual analysis were noted. As time passed, more phoneme–grapheme conversion errors were observed, reflecting MB's attempt to deal with the complex sound–spelling patterns of English.

Our subject, DS, differed from MB in a fundamental way as her reading performance was relatively accurate even immediately after the stroke. Measures of accuracy were thus redundant and, instead, we tracked the evolution of her pure alexia using computer presentation of stimuli and on-line reaction time paradigms. Like MB, our subject, DS, exhibited signs of explicit serial letter encoding in the initial stages poststroke. This form of acquired dyslexia, in which individual letters are slowly and sequentially sounded out, has been termed spelling dyslexia (Kinsbourne & Warrington, 1962), word-form dyslexia (Warrington & Shallice, 1980), and more recently, letter-by-letter reading (Patterson & Kay, 1982). The plethora of labels reflects the numerous theories surrounding this deficit.

Before the various explanations put forward to account for this reading impairment can be examined, the cardinal symptoms of the disorder must be specified. The hallmark of this reading disorder is a word length effect whereby words are identified through their constituent parts and response latency is a monotonic function of the length of the stimulus array. Although time is the critical feature in letter-by-letter reading, no firm criteria or time boundaries have been set down. Reading times ranging from 1.8 to 16.9 sec for a 3- to 4-letter word and from 3.0 to 47.5 sec for a 7- to 8-letter word have been reported for different letter-by-letter readers (Patterson & Kay, 1982; Warrington & Shallice, 1980). Although there is much variability in the range of times observed in these patients, even the fastest times reported are considerably slower than the 400 msec required by a normal adult reader to read a common word out loud (Rayner & Pollatsek, 1989).

Despite the fact that the symptoms of letter-by-letter reading are relatively well accepted, there is little consensus on the mechanism responsible for the impairment and lesions originating in peripheral as well as more central reading processes have been implicated. One explanation for the disorder suggests that letter-by-letter readers have a fundamental problem in perceptual analysis; consequently, the encoding of single letters is abnormally difficult and gives rise to elevated perceptual thresholds (Friedman & Alexander 1984). An alternative account suggests that the problem is not in the recognition of single items but rather in the synthesis or integration of multiple visual forms. This more general visual disturbance, termed simultanagnosia, manifests as the reduced ability to apprehend separate visual forms, leading to individual item analysis (Coslett & Saffran, 1989a; Kinsbourne & Warrington, 1962). According to Warrington & Shallice (1980), the deficit in the automatic synthesis of individual units is not general, but is produced by a primary impairment in the word-form system in which the orthographic representations of previously encountered words are stored. Because the representations of words are affected, their perceptual status is relegated to that of nonwords and they are no longer rapidly identified. Variables such as concreteness or word frequency do not influence performance and words cannot be comprehended unless the individual constituents are processed in this sequential manner. Since the permanent description of words cannot be activated, reading is mediated by an auditory spelling mechanism which accepts letter names as input. Thus, the overt strategy of letter-by-letter reading "is an attempt to *compensate* for damage to a stage in the reading process prior to phonological or semantic analysis" (Warrington & Shallice, 1980, p. 108). Patterson and Kay (1982) refute the claim that the deficit is in the word-form system itself and provide empirical evidence from their letter-by-letter reader that the individual graphemic analyzers and the word-form system are both intact. Their results show that the deficit lies somewhere between these two systems, disconnecting them in the way suggested by Dejerine (1892) in his classical interpretation of alexia without agraphia. The disconnection manifests as an inability to map abstract letter identities in parallel onto existing word-level representations.

Support for the view that the word-form is not destroyed is provided by the observation that some letter-by-letter readers are capable of making correct semantic judgments even when the target word has been presented at intervals too brief for explicit identification (Coslett & Saffran, 1989b; Shallice & Saffran, 1986). In other words, whole-word codes are preserved and can assist in determining the identity of the stimulus item. Bub, Black, and Howell (1989) have shown that, in addition to semantic knowledge, higher-level orthographic influences may be preserved in these subjects. Adopting a paradigm which has been used in normal subjects to demonstrate the superiority of words relative to nonwords (Reicher, 1969; Wheeler, 1970), they showed that their letter-by-letter, JV, reveals better performance for words relative to nonwords in a forced choice task in which he has to decide which of two probed letters appeared in the original briefly exposed target. This influence of orthographic context, presumably the result of activating higher-level codes however, does not operate very strongly in a free report task in which the subject names the letters in the target, suggesting that the existing orthographic information may not be sufficiently detailed or activated for an overt response.

The diverse explanations cited above suggest that letter-by-letter reading is an overt behavior which may be produced by a deficit at any one of a number of stages in the reading process. Like the regularization errors in surface dyslexia which may be the result of a deficit in any one of seven locations (Coltheart & Funnell, 1987), letter-by-letter reading could also arise because of deficits at several possible lesion sites. The laborious encoding strategy could be an adaptive strategy arising subsequent to damage at very early stages of reading (Reuter-Lorenz & Brunn, 1990; Farah & Wallace, 1990) or at later stages (Warrington & Shallice, 1980) or at any one of a number of places between the two (Kay & Hanley, 1990; Patterson & Kay, 1982). The goals of our study, therefore, were to document the site of the underlying deficit in our subject and to obtain fine-grained details of her letter-by-letter reading. We then aimed to take measurements over time and to compare them to the baseline data to determine whether any recovery took place. We hypothesized that any recovery might be the product of a change in the underlying deficit itself, or alternatively, that the underlying impairment may not resolve but that the use of the adaptive strategy may become more efficient over time. This distinction is important. In the first case, reading would be more similar to the normal process whereas, in the latter, the disorder would persist but would be obscured by better use of the alternative, abnormal behavior. To differentiate between these two possible sources, we aimed to reexamine the competence of the reading system at one year poststroke to determine whether the underlying deficit had resolved.

Although letter-by-letter reading has been described in some detail in the literature to date, the interest has been on the nature of the disorder rather than its change over time (Bub et al., 1989; Coslett & Saffran, 1989b; Friedman & Alexander, 1984). Although Friedman and Alexander (1984) tested their letter-by-letter reader over a 2-year period, their focus was on the residual deficit. They provide few details of its evolution, except for remarking that at 22 months postsurgery "both reading accuracy and reading speed have improved at a slow but steady rate" (p. 13), and that "reading has not yet achieved automaticity." Since recovery has important implications for remediation which attempts to exploit the natural pattern of recovery and since little is known about the progression of acquired dyslexia, we evaluated the performance of our pure alexic subject over several months using chronometric measures and employing reaction time as an index of recovery. Longitudinal data of this type have not been reported in the literature and they may contribute to our understanding of the underlying disorder.

CASE REPORT

DS, a 32-year-old right-handed nurse who was born in Bombay was admitted to hospital in November 1986 with sudden onset of left occipital headache and dizziness. DS's mother tongue was English and although she had spoken the vernacular during her childhood in India, she was schooled in English and speaks it exclusively at present. On initial examination, she had a right homonyous hemianopsia and mild right hemiparesis. On four-vessel angiography, she had an occlusion of the posterior cerebral artery and a corresponding left occipital infarction was seen on CT scanning. A migrainous etiology was inferred in view of a previous and subsequent history of migraine headache and by exclusion of other possible contributing factors by extensive investigation.

On detailed assessment of her language function shortly after her stroke, her primary difficulty was with reading. She used a sequential letter-by-letter procedure to decipher words. Her auditory comprehension and spoken language production were good. Occasional word-finding difficulty detected on the Boston Naming Test was attributed to cultural bias; for example, she could not name hammock or pretzel. DS's hemiparesis resolved quickly and she was discharged home a few weeks after admission. She continued on an out-patient occupational therapy program which focused on activities of daily living and homemaking.

Visual field testing by Goldman perimetry shortly after her stroke revealed a congruous right homonymous field defect, denser in the superior quadrant. This had improved to a right upper quadrantanopsia at 8 months follow-up. Neuropsychological evaluation 8 months after her stroke revealed deficits in attention and concentration, in verbal learning and recall, and in some visuospatial functions. Her verbal IQ (88) fell into the lower 25th percentile and her performance IQ (74) in the lower 5th percentile on the Wechsler Adult Intelligence Scale Revised. Performance on the Wechsler Memory Scale (83) was commensurate with her full-scale IQ (80). Again, there were no aphasic deficits noted and reading comprehension was normal although reading was noted to be slow. A reevaluation 18 months after onset showed improvement in her performance IQ (89), which now fell in the lower 25th percentile. Her verbal IQ (91) and her full scale score (89) showed a minor improvement. Although DS continued to show mild impairments in attention and concentration, verbal learning, and recall, she had resumed her normal lifestyle and her occupation as a homemaker. She could read children's books to her two children but found everyday reading laborious and unenjoyable.

Lesion Localization

A CT scan repeated 3 months postonset (see Figs. 1a, b, and c) was unchanged from the admission scan except that the hypodensity in the occipital region was more clearly defined. It involved the medial occipital temporal region including the fusiform, lingual, and cuneate gyrus, the calcarine cortex, and the hippocampus. Thus, Brodmann's areas 17, 18, 28, 19, and 37 were partially affected by the lesion. The hypodense area also extended into the occipital white matter including the optic radiations and patchy involvement of the forceps major of the splenium. It is noteworthy that a lesion involving the inferior paraventricular white matter was also found because this area is considered critical for the development of pure alexia (Damasio & Damasio, 1983). The posterior lateral thalamus and posterior internal capsule also appeared to be partially affected.

TESTING

The testing was divided into three stages: documenting the letter-by-letter pattern shortly after the onset of the stroke, locating the underlying site of the lesion, and, finally, evaluating the changes in performance over time.

A. Documenting DS's Reading Behavior

These tests, conducted within the first week poststroke, were designed to characterize the severity of the initial deficit and to set the baseline for later comparison. Single words were presented to DS's intact left visual field using an Apple IIE computer and monitor linked to a millisecond reaction time clock. DS sat at a comfortable distance of 50 cm from the monitor. The computer presentation consisted of a central fixation point which remained on the screen for 1 sec and was followed, after a delay of 500 msec, by the target stimulus which subtended a vertical angle of 0.5°. Reaction times as well as accuracy scores were collected. This general procedure was also used for subsequent testing.

For the single word testing, a list of 120 uppercase words, 40 three letters, 40 five letters, and 40 seven letters in length, were presented individually following the central fixation point. The mean frequency of the words was controlled across lengths (mean 52, SD 70). Half the list contained low-frequency words appearing less often than 20 times per million and the other half contained high-frequency words, appearing more than 20 times per million (Kučera & Francis, 1967). Half the list consisted of abstract words and the other half, concrete words. All the words had regular sound–spelling correspondences and there was one homophone (sum).

The words appeared for unlimited exposure duration until DS responded by activating a voice relay key or by depressing a response key depending on the task (see below). The stimuli were right-aligned so that the final character of all words fell in the space immediately adjacent to fixation. The visual angles subtended were 1.5°, 2.4°, and 3.6° for 3-,

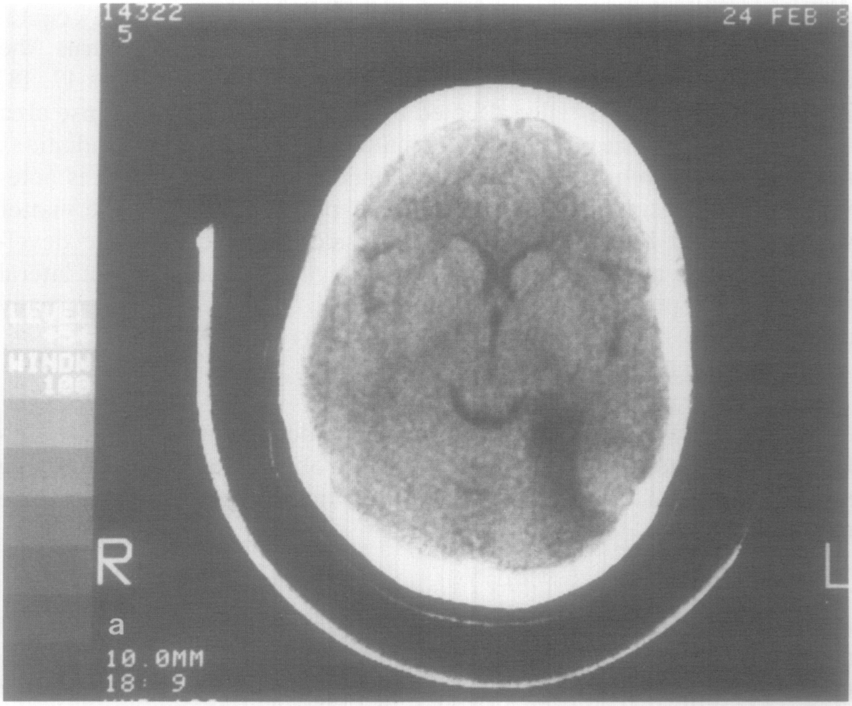
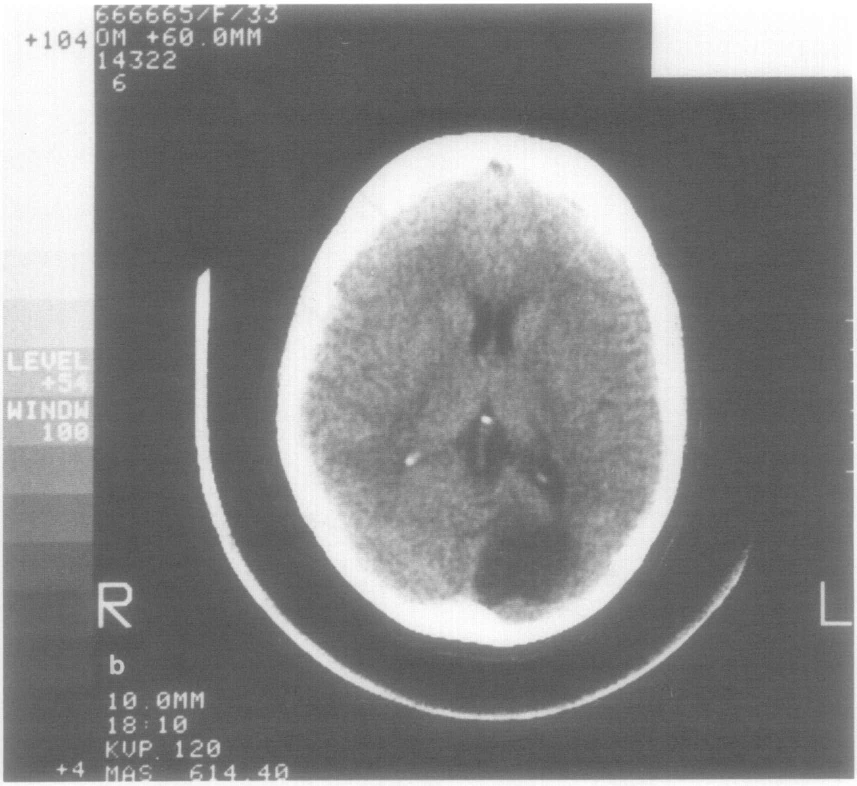


FIG. 1. Axial C.T. scan at 15° to orbitomeatal line obtained on a GE9800, reveals a left occipital infarction. The details of localization are discussed in the text.

5-, and 7-letter words, respectively. A 3-sec delay, measured from response time, preceded the following trial.

1. Oral reading. DS was instructed to read the words aloud as quickly and as accurately as possible. A microphone-activated voice relay mechanism was used to measure reaction time and accuracy was recorded manually. A set of practice trials was administered. DS read 100/120 words correctly. We noted that she named each individual letter out loud before pronouncing the whole word. The four errors all arose from letter misidentifications, e.g., joy → "jay." There was no significant effect of frequency (Fisher exact test high vs. low frequency, $p = .14$) nor word type (Fisher exact test concrete versus abstract $p = .36$) on her oral reading.

2. Lexical decision. The 120 words described above were combined with 120 orthographically legal and pronounceable nonwords (for example, foy, trisk) derived by changing 1 or 2 letters of the companion real word. The stimuli were presented individually following the fixation point and DS was instructed to use her dominant hand to depress the "yes" key for words and the "no" key for nonwords. Practice trials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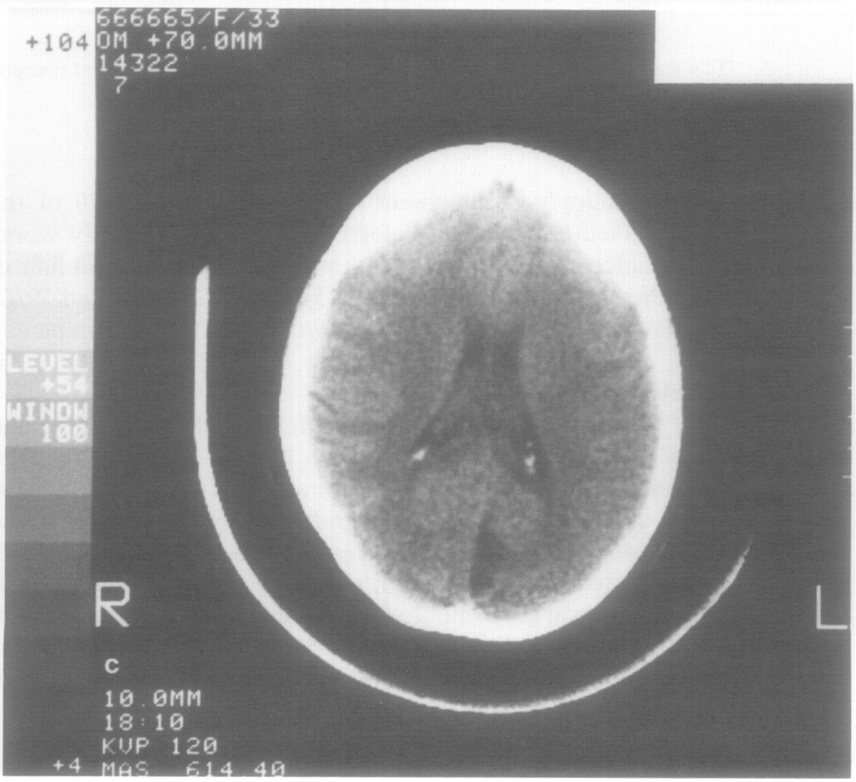
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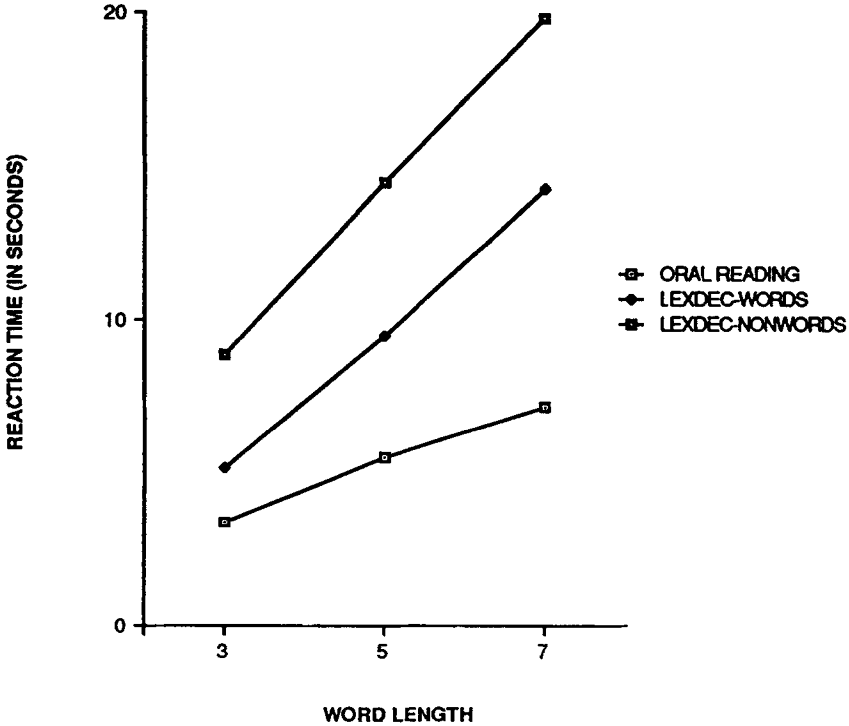


FIG. 2. DS's mean reaction time per word length on oral reading and lexical decision one week postonset.

were provided. Accuracy of judgment was high, with 110/120 of the words and 102/120 of the nonwords correctly classified, slightly worse than the performance of a group of 10 normal subjects who read half of the items from the same task and scored 59.5/60 on the words and 59/60 on the nonwords. As on oral reading, there were no effects of frequency nor word type on DS's performance (Fisher exact test $p = .08$, $p = .18$, respectively).

The average response times per word length for both oral reading and naming latency are illustrated in Fig. 2.

Speed of reading is obviously very slow, as reflected in the long latencies on both tasks. A significant linear relationship between time and word length was demonstrated ($r = .99$, $p < .05$ oral reading; $r = .98$, $p < .05$ for lexical decision real words), reflecting a strong length effect. The longer times for nonwords relative to words on lexical decision are consistent with the findings that nonwords have different status in reading (Rayner & Pollatsek, 1989). Nonwords, therefore, served only as foils and were not analyzed further.

DS required, on average, 1.2 sec per letter in oral reading and 1.8 sec per letter in lexical decision. These times place her approximately in the middle of the severity range set by Shallice (1988) in which the least severe letter-by-letter reader required 1.8 sec for a 3- to 4-letter word and the most severe required 16.9 sec.

B. Locating the Underlying Impairment

Several stages of reading were investigated in an attempt to locate the functional lesion giving rise to the letter-by-letter reading.

1. Single letter analysis. A list of 60 single upper case letters was presented for letter naming. Each letter appeared individually for the briefest possible exposure duration (subject to screen refresh time and phosphor decay on the Apple IIE monitor) following a central fixation point. DS was able to identify 58 of the 60 letters even at this brief duration, suggesting that her letter-by-letter reading could not be attributed to a perceptual failure in recognizing individual items.

2. Integrating multiple items. Two- and three-item letter arrays (a single character space between letters) were also presented to DS in the same way as the single items described above. She recognized 45/60 items of the two-item arrays at 32 msec and 47/60 of the three-item arrays at 48 msec. Thus, although she was able to identify the multiple item arrays correctly at unlimited exposure durations, she did experience difficulty integrating separate units at very brief exposures.

3. Lexical access. In order to determine whether DS, like other subjects (Coslett & Saffran, 1989b; Shallice & Saffran, 1986), retained implicit or unconscious knowledge about words which were presented too briefly for explicit report, we constructed a semantic classification task which we presented at limited exposure duration. The fact that, in some subjects, implicit knowledge is preserved although not necessarily utilized in explicit report tasks is theoretically significant. Dejerine's (1892) original theory proposed a total visual–verbal disconnection, thus preventing any information from reaching the left hemisphere language area. The fact that visual information may be gaining access to verbal mediation argues against this view and, instead, is more consistent with the recent notion that the right hemisphere may be responsible for lexical access in these patients (Coslett & Saffran, 1989b).

Our task involved the presentation of three lists of 60 words (5–7 letters in length) using an exposure duration (3 sec) at which DS would be unable to read the entire word explicitly. She was instructed to report the word if possible or if she could not read it, to judge (or guess if necessary) whether the word was related to work or leisure (list 1, e.g., tennis, salary), whether it was associated with positive or negative emotion (list 2, e.g., malice, honesty), or whether it referred to an animal or object (list 3, e.g., beaver, pencil). DS was not significantly different from chance (50%) in her judgments of those words ($n = 118$) she could not identify explicitly ($\chi^2 = .34$, $p > .10$), suggesting an absence of preserved implicit semantic knowledge.

4. Orthographic context effects. In order to assess whether DS was able to synthesize any higher-level orthographic units from perception, a finding which would favor adequate access and use of the word-form, we adopted a paradigm which has been used extensively with normal subjects (McClelland, 1976; McClelland & Johnston, 1977) and more recently with subjects with acquired dyslexia (Bub et al., 1989; Kay & Hanley, 1990; Reuter-Lorenz & Brunn, 1990). The word superiority effect (WSE), in which a letter in a real word is recognized more quickly than a letter in a nonword, which, in turn is recognized more quickly than a letter in a random string, is taken as evidence that higher-level orthographic representations play a role in perception. While whole-word activation may account for the former, the activation of subword units is thought to account for the superiority of pseudowords over random strings. Bub et al. (1989) have shown that their letter-by-letter reader, JV, was able to recognize more letters in words and pseudowords than in nonwords in all positions of a 4-letter display when a forced choice procedure was employed. In a free report task, in which the subject was instructed to report all the letters, however, his performance was significantly poorer in the last two positions compared to the first two positions of the array. The fact that a WSE was observed, albeit only on the forced choice task, led the authors to conclude that whole-word codes are preserved but that the activation from print, especially in the final positions, is weak and may not be available for explicit naming in the free report task.

We used the same forced choice procedure as Bub et al. (1989) to assess whether DS could activate higher-level codes from print. We predicted that if she was able to use the intact word representations and/or orthographic knowledge, she would be able to report more letters in words than in pseudowords and would show poorest performance on random strings irrespective of letter position in the array.

Three groups of 40 four-letter strings were obtained, one of high-frequency words (more than 55 per million), one of pseudowords, and one of random strings. The pseudowords were matched to the real words in bigram frequency and the letter sequences all had a zero bigram frequency. Each string was presented in DS's intact visual field for a limited exposure duration following a central fixation point. A random pattern mask followed for 500 msec. The target duration was set at 450 msec, the exposure at which DS scored 70% accuracy at letter detection in pseudowords. The relevant measure, then, was the accuracy of letter detection as a function of the different target types. The forced choice probe was conducted in two ways:

a. Whole-word condition. Two 4-letter strings appeared after the mask, one of which was the target item and the other a distractor, differing from the target by a single letter. The different letter appeared equally often in each of the four letter positions. In the case of words, the distractor was also an English word; for example, when the target was BOOK, the distractor was BOOT. The position of the target (either left or right) was counterbalanced across trials. The task was blocked with letter strings being presented first, followed by pseudowords and then words.

b. Single-letter condition. Two single letters appeared after the string, with their relative word position marked, for example, for the target BOOK, the probes were ---T or ---K. Each letter position was probed an equal number of times and in the case of words, the distractor letter could also complete an English word as in the example provided above.

The numbers of correct letter detections on both whole-word and single letter probes are shown in Fig. 3.

There was no significant difference between DS's ability to detect letters in words relative to pseudowords or random strings on either the whole-word trials (χ^2 = 1.39, p = .5) or on the single-letter trials (χ^2 = .8, $p > .5$). There was, however, a significant difference in her ability to detect the target letter in the first two positions of the array compared to the last two positions (positions 1 and 2 vs. positions 3 and 4, collapsed across target types, χ^2 = 10.67, $p < .01$). These findings suggest that DS was not able to use higher-order knowledge to assist in the perceptual synthesis of the 4-letter displays on either procedure. The results are consistent with the findings of Kay and Hanley (1990) whose subject, PD, showed no word superiority and who was unable to identify more than one letter presented simultaneously. While DS had sufficient time to encode the first two letters serially, proceeding from left to right, leading to better performance in these positions, the exposures were too brief to allow sequential encoding of all four positions.

In sum, DS demonstrated, in the acute stage of her illness, the classical symptoms of a severe letter-by-letter reader with relatively intact peripheral and visual analysis. While she was able to access higher units of lexical semantic information when the target appeared for unlimited duration (leading to high accuracy scores in the lexical decision and oral reading tasks), she was unable to access this information rapidly when the target appeared too briefly for explicit encoding of the component letters.

C. Longitudinal Testing

Following the preliminary testing, DS was assessed on 10 subsequent occasions over a 12-month period. The oral reading and lexical decision tasks described above were administered on each occasion. An additional

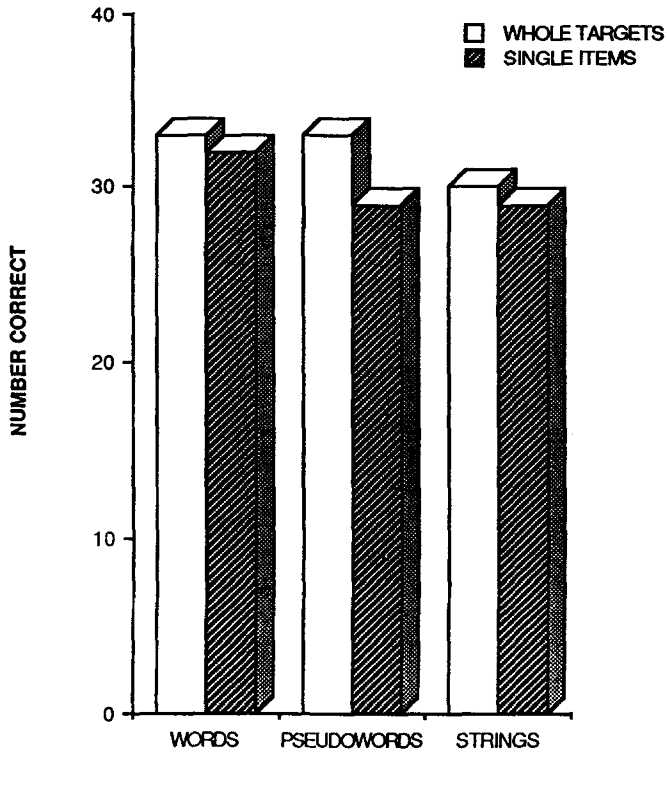
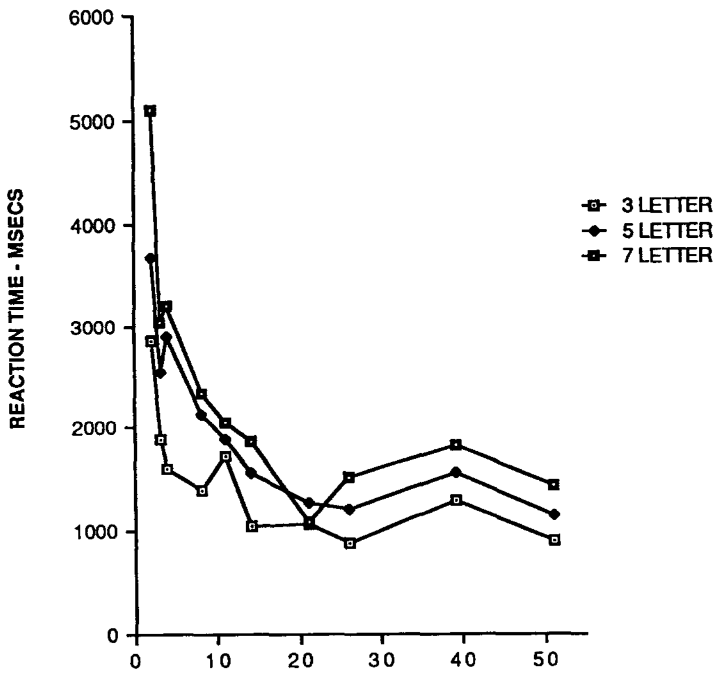


FIG. 3. Number letters correctly detected in words, pseudowords, and random strings on a forced-choice task.

five lists of words were compiled (and intermixed with nonwords for lexical decision) using the same criteria for length, frequency, and word type as the original list. The lists were presented in random order over time. The administration procedure mirrored that of the preliminary testing and accuracy and latency measures were obtained in each session for oral reading and for lexical decision.

1. Oral reading. DS's accuracy in oral reading (expressed as percentage errors) was consistently high across all time periods and all word lengths, ranging between 92 and 100% correct. Reaction times in oral reading, expressed as a function of time since onset, are illustrated in Fig. 4.

A repeated measures analysis of variance with time since onset as a between factor and word length as a within factor was conducted. There was a significant main effect of time ($F(9, 18) = 19.1$, $p < .001$), with reaction time decreasing with an increase in number of weeks since onset



TIME POST-ONSET (WEEKS)

FIG. 4. DS's mean reaction time in oral reading as a function of time period.

of the stroke. A significant effect of word length was also noted ($F(2, 18) = 4.89$, $p < .05$), with time for 3-letter words (1635.1. msec) being significantly faster than those for 5-letter words (1983.3 msec) which, in turn, were faster than those for 7-letter words (2340 msec). The recovery curves for each word length, plotted as a function of time since onset, showed the best fit to curvilinear regression lines ($r = 0.8$ for 3 letters, 0.9 for 5 letters, and 0.85 for 7 letters), with negative deceleration from 30 weeks postonset. In addition, a significant linear relationship between word length and response latency, with an increase in processing time with every additional letter, was observed in each individual testing session with the exception of 21 weeks postonset ($r = .09$) at which point, the 7-letter words converged on the 3-letter words. The regression slopes and linear trends plotted for word length on reaction time for the 10 testing sessions are presented in Appendix I.

In the final testing session, at 51 weeks postonset, DS's mean reading times of 896.3, 1138, and 1442 msec for 3-, 5-, and 7-letter words, respectively, showed a significant linear trend ($r = 1.00$) with an intercept of 612.3 and a slope of 273 msec for each additional letter. These times

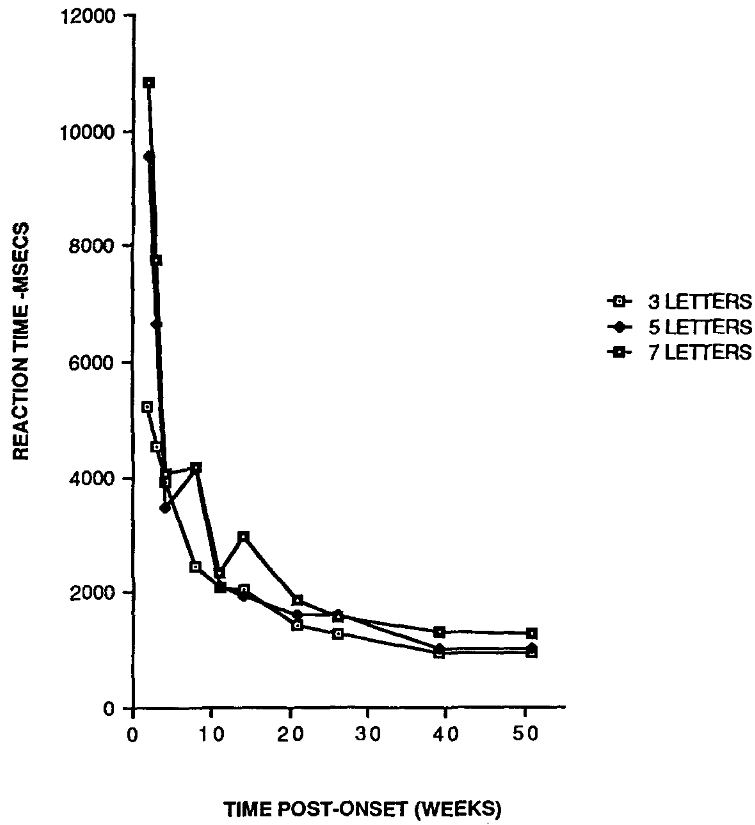


FIG. 5. DS's mean reaction time on lexical decision as a function of time period.

far exceed the slopes of 30 msec per letter found in skilled readers when a stimulus word is presented in the left visual field (Bub & Lewine, 1988) but are more consistent with the 1–1.3 sec per letter time of JV, another letter-by-letter reader (Bub et al., 1989). These findings suggest that although there has been a dramatic improvement in response latency as a function of time since the stroke, DS still shows elevated reaction times and the length effect which is characteristic of letter-by-letter reading.

2. *Lexical decision.* As on oral reading, the accuracy scores on lexical decision were consistently high, ranging between 90 and 98% correct for "yes" responses. The reaction times, as a function of testing period, are illustrated in Fig. 5.

An analysis of variance with time as a between factor and word length as a within factor was conducted. The results reveal a significant effect of testing session on reaction time ($F(9, 18) = 18.8$, $p < .001$) and a significant effect of word length ($F(2, 18) = 5.3$, $p < .05$). The mean reaction times per word length (collapsed across testing sessions) were 2486.5, 3317.6, and 3815.2 for 3-, 5-, and 7-letter words, respectively.

The data were best fit to a curvilinear regression line demonstrating rapid decrease in response latency up to 20 weeks postonset with more gradual decrease thereafter. The monotonic relationship between word length and reaction time was observed in all but one testing session (4 weeks postonset) as seen on the regression slopes and linear trends (see Appendix I). The negative deceleration in latency is confirmed by the relatively small diffference in the number of seconds taken to read a single letter at 39 weeks postonset compared to the number of seconds required at 51 weeks postonset. Observation of DS over an additional 4 months (up to 75 weeks) revealed no major changes in her performance, providing further support for the asymptote we documented.

The results of the lexical decision task, then, like the oral reading task, demonstrate rapid decrement in response latency over time, with performance approaching an asymptote in the final periods. In order to obtain an estimate of normal reading speeds on oral reading and lexical decision, we administered our tasks using the identical administration procedure to a normal control subject, matched to DS in age, sex, and level of education. Mean reaction times on oral reading were 589, 551, and 579 msec for 3-, 5-, and 7-letter words, revealing a nonsignificant regression slope of 4 msec ($r = .63$, $p > .05$). Mean reaction times for lexical decision were 497, 505, and 553 msec for 3-, 5-, and 7-letter words, respectively, producing a nonsignificant regression slope of 11 msec for every additional letter ($r = .77$, $p > .05$). It is clear that DS's mean reaction times were considerably slower than those of a matched control subject and that, unlike the normal control, she continued to show a linear trend in her data even in the final testing session.

3. Orthographic context effects. At one year postonset, we reexamined the nature of the underlying reading deficit in DS to determine whether the changes observed in her reading performance might be attributable to her being able to gain rapid access to the visual word-form system. Instead of repeating the forced-choice probe procedure employed initially, we utilized a free-report paradigm which, according to Bub, Black, and Behrmann (1990), is a more sensitive measure of whole-word activation. The use of a binary response in the forced-choice procedure renders the task relatively insensitive because the level of accuracy, by chance alone, is 50%. Guesswork in the free report measure, in which the subject names the perceived letters, is minimized by using target items whose identity cannot be guessed on the basis of three letters.

In the following task, we used this free-report paradigm to assess DS's ability to access the word-form system. The strongest prediction was that if the change in reading performance were attributable to the resolution of the primary deficit, then we might expect to see the emergence of a word-superiority effect (WSE). If however, there was no recovery of this fundamental inability to rapidly access word forms, then no WSE would be evident and the changes in DS's reading performance could not be attributed to the resolution of the underlying deficit.

A set of 4-letter words ($n = 120$) was used (taken from Bub, Black, & Behrmann, 1990). All words yielded at least two plausible responses if completed on the basis of their first three letters; for example, the target SEEN could be completed as SEEM, SEEK, SEED. Nonsense words whose summed bigram frequencies were either equal to or greater than the counterpart word were formed by changing one letter of the target. In this task, DS was instructed to report the letters without guessing the identity of the unseen letters. The targets were presented to her intact left field and followed by a random pattern mask for 500 msec. The targets were all presented at 150 msec, the exposure duration at which DS obtained 70% correct letter detection in pseudowords. The presentations of word and pseudoword targets were blocked. Two measures were taken: the total number of letters detected correctly from the target and the total number of full words (all four letters) correctly reported. Using this measure, Bub, Black, and Behrmann (1990) found that normal readers showed a clear difference (35%) between words and pseudowords, when the number of whole-words perceived correctly is measured. The difference between the words and pseudowords was confirmed on an item analysis which showed a significant difference across all items ($t(119) = 14.01$, $p < .001$). According to these authors, this technique provides a sensitive measure of a word-form deficit, indicating whether or not the lexical representations are accessed and utilized. The results of these measures for words and nonwords are shown in Fig. 6.

As is evident from this figure, there is no overall significant difference in DS's ability to detect letters or entire targets on words compared to pseudowords on either of the two measures; i.e., there is no overall WSE (Fisher exact test collapsed across all frequencies; letters $\chi^2 = .07$, $p > .5$; entire targets $\chi^2 = 5.13$, $p > .5$). The mean single letters correctly reported were 76.1% for words and 75% for nonwords while the mean whole words correctly reported were 43 and 39.5% for words and nonwords, respectively. The absence of a WSE suggests that even at one year postonset, DS was still unable to use higher-level orthographic context to assist in perception.

DISCUSSION

We have described the evolution of a reading deficit over a 12-month period of a subject with pure alexia, largely unaccompanied by any other significant neurobehavioral deficit. In the first week following the stroke, DS read laboriously and sequentially and was classifiable as a letter-by-letter reader of moderate severity according to the continuum of reading

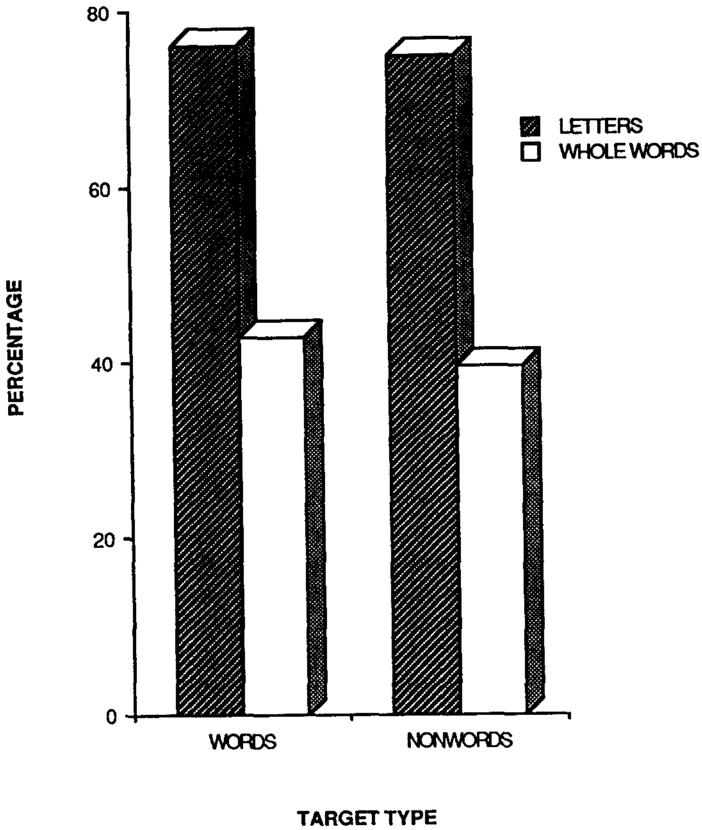


FIG. 6. Percentage letters correctly detected in words and pseudowords on a full-report task at one year poststroke.

times set by Shallice (1988). DS was able to recognize letters accurately, provided that enough time was given for explicit encoding. Her poorer performance on multiple-item arrays at brief exposure durations suggests an impairment in rapidly identifying several items presented simultaneously (see Kay & Hanley 1990, for a similar case). DS was able to encode the first two letters of a 4-letter array reliably but was not able to process the final two letters under brief exposure durations. The absence of a word superiority effect reflects the inability to access higher-order orthographic structure for detecting letters in familiar words, pseudowords, and random strings.

Over time, DS showed remarkable improvement in her reading performance: her reaction times in oral reading and lexical decision tasks decreased dramatically although, even in the final testing session, her reading times were still considerably longer than those reported for normal subjects. DS's reading times were also slower than the times reported for a group of nonalexic left-occipital damaged patients tested on the same tasks with the same materials (Black, Bub, & Behrmann, 1987). Mean reaction times for a group of five patients with single left inferior occipital lesions for 3-, 5-, and 7-letter words, respectively, were 860, 895, and 950 msec (DS at 51 weeks: 896.3, 1138, 1442) on oral reading and 884, 887, and 1003 msec (DS at 51 weeks 1303, 1555, 1712 msec) on lexical decision. The mean rate of processing in this group was 30 msec per letter, which is far less than the 168.5 msec (lexical decision) or 273 msec (oral reading) per letter required by DS in the final testing session. DS's performance is clearly abnormal even as it approaches an asymptotic level. In addition, she still showed an effect of word length on response latency. This persisting monotonic relationship between length and time suggests that she was still a letter-by-letter reader.

In view of the fact that the alteration in reading performance appears to be more quantitative than qualitative, to what underlying mechanism can we attribute the difference? A reanalysis of the word-form deficit still failed to reveal a word superiority effect even when a more sensitive free-report procedure was used in place of the forced-choice task. The absence of higher-order contextual influences suggests that access to the word-form had not improved and the observed changes in DS's performance were therefore unlikely to be the result of the recovery of the primary deficit. Rather, we infer that her improvement reflected increased efficiency in the use of sequential letter-by-letter spelling as a form of compensation. Thus, although DS still seemed to be processing letters serially rather than in parallel, increased speed in naming individual letters or in exploiting knowledge of orthotactic patterns may well have led to the improvement in her reaction times In other words, she seemed to have refined her compensatory procedure but had still not achieved automatic access to the word-form system. Support for this interpretation is obtained from another letter-by-letter reader (Friedman & Alexander, 1984) who, despite changes in behavior over a 2-year period, had still not achieved automaticity and was still defined as a letter-by-letter reader.

An important question which arises from these findings concerns the way in which automaticity may be achieved. One possible avenue for reacquisition is through the mediation of this sequential letter-by-letter strategy. Whether such an identification procedure is an adequate means for ensuring access to the visual word-form, however, is unclear. Some data from the childhood developmental literature suggest that it is. Seymour and Porpodas (1980), for example, have shown that a word length effect is a critical dimension in reading acquisition. They described word lengths in the order of 200 msec per letter in a group of normal 7-year-olds. This length effect seemed to be critically involved in the process of constructing word representations. As reading skills were refined with age and word forms were stabilized, so this length effect appeared to decrease. It is questionable, however, whether the laborious sequential reading process utilized in letter-by-letter reading facilitates the reacquisition of word forms or not. The fact that DS's performance was approaching an asymptote at levels well beyond the normal times (and beyond the usual period of spontaneous recovery) suggests that this might not be the case.

The recovery curve mapped out in our subject has important implications for remediation. An alternative, wholistic approach might be necessary to ensure rapid access to the word-form system since the adopted compensatory strategy does not seem to suffice. Intervention of this sort, of course, is possible only if letter-by-letter readers are detected clinically and correctly diagnosed. This is often not the case. In fact, it has been suggested that letter-by-letter reading is not well recognized in a clinical setting for a number of reasons. It is relatively rare in its pure form, conspicuous neurobehavioral problems are absent, and the loss of the reading ability is often not considered limiting (Marks & De Vito, 1987). Indeed, if DS were to be seen in a clinical setting at one year postonset, it is unlikely that her dyslexia would have been discovered on routine clinical testing, as reading accuracy was good. The overall inefficiency of her reading ability, however, continued to take its toll, making reading burdensome. Chronometric measures such as those described in this paper may therefore serve a useful diagnostic purpose indicating the nature of the reading disturbance and documenting the changes in performance during the recovery process.

APPENDIX I

Regression Slopes and Linear Trends for Oral Reading and Lexical Decision in Relation to Time Postonset

Time postonset (weeks)	Oral reading Slope (msec)	Oral reading Linear trend *r*	Lexical decision Slope (msec)	Lexical decision Linear trend *r*
2	1128	0.99	2795	0.98
3	575	1.00	1607	0.98
4	792.5	0.94	72	0.23
8	477	0.95	861.5	0.87
11	162	1.00	445	0.81
14	405.5	0.99	136.5	0.96
21	10	0.09	218.5	1.00
26	316	1.00	160.5	0.81
39	263	1.00	179	0.93
51	273	1.00	168.5	0.94

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