

Pure alexia is a well-documented syndrome characterized by impaired reading in the context of relatively intact spelling, resulting from lesions of the left temporo-occipital region (Coltheart, 1998). It is considered a disconnection syndrome in that visual information about written words does not reach the cortical areas critical for word recognition (i.e., the orthographic lexicon). The relative preservation of orthographic knowledge is confirmed in individuals with pure alexia by the fact that they can spell and they can also recognize orally spelled words with little difficulty. To compensate for their deficit, pure alexics often use a letter-by-letter (LBL) strategy, where reading is achieved by sequentially identifying the component letters of a word. In some cases, however, letter identification is impaired and a LBL reading approach is obviated. The inability to name letters may reflect a visual perceptual deficit, or a disconnection of visual-verbal information, where the letters are correctly perceived but fail to access letter names.

The treatment of pure alexia characterized by poor letter-naming is particularly challenging. Several investigators have used a motor/kinesthetic approach to improve letter identification in such individuals (see Table 1 for details; Greenwald & Gonzalez-Rothi, 1998; Lott & Friedman, 1999; Lott et al., 1994; Maher et al., 1998). The rationale for this method is that the orthographic lexicon is accessed through the tactile/kinesthetic modality, circumventing the impaired access through the visual modality. Once letter identification has reached an acceptable level, training is geared toward improving speed and accuracy of reading. In the present study, we report on an individual with severe alexia who appeared to be similar to the cases described in Table 1, and thus, an appropriate candidate for the motor/kinesthetic approach to improve letter identification and single word reading.

Method

Patient Description

TS was a 74-year old, right-handed female with 12 years of education, who was 15 months post onset of a temporo-parieto-occipital stroke at the time of this study. CT scan showed a lesion affecting the inferior and lateral aspects of BA 20/21 and 37, including the “visual word form area.” Superiorly, the lesion extended into the angular gyrus (BA 39) and dorso-medial occipital cortex (BA 18, 19). Extensive damage to interhemispheric callosal fibers traveling in the forceps major was also evident (Figure 1). The stroke resulted in right homonymous hemianopia, anomia, and alexia with agraphia. Her spoken language profile was consistent with anomic aphasia, with an Aphasia Quotient of 77 on the *Western Aphasia Battery* (WAB; Kertesz, 1982).

Pre-treatment Assessment

TS' reading and spelling performance was assessed using a list of 40 regular and 40 irregular words (balanced for frequency, length and imageability), and 20 pronounceable non-words. She demonstrated significantly impaired reading (12% correct overall) relative to recognition of oral spelling (65% correct overall). Her reading was characterized by attempts to read letter-by-letter, but with frequent errors in letter identification. Writing words to dictation was better preserved than reading, but was also impaired (76% correct overall). Her spelling of irregular words was more impaired than

spelling of regular words and nonwords, a profile consistent with lexical agraphia. It was noteworthy, however, that TS was able to write words to dictation with much greater accuracy than she was able to copy printed words (see Figure 2).

Additional tests of visual/orthographic knowledge and peripheral writing abilities showed marked deficits in copying single words, case conversion (e.g., writing uppercase letter in response to visual presentation of lowercase), and naming visually-presented letters (see Table 2). In contrast, TS was able to correctly perform visual processing tasks including the identification of mirror-reversed letters, and matching of upper-to-lowercase letters, and vice versa. She was also able to correctly write all letters of the alphabet to dictation, write the letters associated with spoken phonemes (18/20), and could write CVC nonwords like “meb” with few errors (57/60 sound-letter correspondences correct). Written word-to-picture matching was surprisingly good (38/40) considering her profound impairment in oral reading of single words. Thus, performance was near normal on purely visual tasks, but impaired on tasks that required links between visual input and spoken or written output modalities.

Treatment

Treatment was aimed at improving reading of single words using a motor/kinesthetic approach to facilitate the recognition of component letters. TS was trained to copy individual words, attempting to name each letter aloud as she wrote it. Correct sequential writing and naming of the letters was expected to result in correct word recognition. Daily homework was provided using high frequency, imageable nouns ranging from 3-5 letters that were trained in sets of six. For each word, a picture and the printed word were provided. TS was instructed to look at the picture, then copy the word attempting to name each letter aloud, and finally say the word. This adaptation of the Copy and Recall Treatment (CART) that has been used to retrain spelling for targeted words (Beeson, 1999), was intended to facilitate reading by providing semantic information from the picture along with lexical information derived using the motor/kinesthetic strategy.

Results

TS received fifteen 1-hour treatment sessions over the course of 8 weeks. As shown in Figure 3, letter naming improved from 12/26 correct at pre-treatment to an average of 25/26 correct over the last three sessions, by employing the motor/kinesthetic strategy to self-cue her responses. Re-administration of the single-word reading list showed a slight improvement in oral reading (12% to 17% correct overall), but this difference was not statistically significant ($\chi^2 = 2.46, p = .12$). TS attempted to read letter-by-letter supported by the motor/kinesthetic strategy; however, it was extremely laborious. She often required several ‘copies’ of a letter before it was correctly named, adding considerable demands on working memory as she tried to decode the written words. In addition, some of TS’s reading errors reflected intrusions of trained words beginning with the same letter or visually similar to the target (e.g. tone → bone).

Discussion

In the case of alexia with agraphia presented here, a motor/kinesthetic strategy of copying individual letters served to improve letter identification, however, improved naming of letters in isolation did not result in a corresponding increase in single-word reading accuracy. This limited response to treatment differs from several other cases reported in the literature. The underlying premise of the motor-kinesthetic approach is that copying of individual letters provides sensorimotor information about letter identities that supports the degraded visual information. It appeared, however, that this cross-modality information was not easily accessible to TS, and that, in addition to a visual-verbal disconnection, TS also had a persistent disruption of visual-motor connections necessary to copy letters. This disconnection was evident on the pre-treatment assessment, but we did not anticipate that it would be resistant to remediation.

The difference between TS and the other treatment cases may relate to the location and extent of the lesion. Although localizing information was somewhat limited in the other cases, it appeared that damage was predominantly in the left inferior temporo-occipital region, as is typical of individuals with pure alexia (Black & Behrmann, 1994). By contrast, TS's lesion had greater dorsal extension and produced more extensive damage to inter-hemispheric white matter fibers. Damage to dorsal callosal pathways is the most likely neural substrate of the visual-verbal and visual-motor disconnections that appeared to underlie TS' persistent reading deficits.

References

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Table 1.

Summary of lesion and selected behavioral characteristics of alexic readers who exhibited positive response to tactile-kinesthetic treatment to improve letter identification

| | Lott, Friedman & Linebaugh 1994 | Lott & Friedman 1999 | Greenwald & Gonzalez Rothi 1998 | Maher et al. 1998 |
|-------------------------------|--|---|---|---|
| Patient | TL | DL | MR | VT |
| Age/Gender | 67/M | 67/M | 72/F | 43/F |
| Time post onset | 14 months | 5 months | 13 months | 14 months |
| Lesion | “residual damage to the left posterior temporal and lateral occipital lobes” | “left posterior temporal-occipital lobe infarct” | “hemorrhagic lesion involving occipital lobe, inferior temporal region to TPO junction” | BA 28, 31, 18, left cerebellar hemisphere |
| Aphasia profile | Transcortical sensory aphasia | Moderate anomic aphasia | Anomic aphasia | “No evidence of aphasia” WAB Naming: 9.7/10 |
| Naming | 56/114 (49% correct) on BDAE confrontation naming | 50/114 (44% correct) on BDAE confrontation naming | 3/60 (5% correct) on BNT | 59/60 (98% correct) on BNT |
| Visual deficit | Lower right homonymous quadrantanopia | None reported | Right homonymous hemianopsia | Right homonymous hemianopsia |
| Oral reading single words | 24/30 (80% correct) on BDAE | 2/30 (7% correct) on BDAE | 0% correct on BARF | 0% correct on BARF |
| Recognition of oral spelling | 8/8 (100% correct) on BDAE | 8/8 (100% correct) on BDAE | 96% correct on BARF | 100% correct on BARF |
| Written spelling | 6/10 (60% correct) on BDAE spelling to dictation | 8/10 (80% correct) on BDAE spelling to dictation | 67% correct on BARF; 77% oral spelling | 100% correct on BARF |
| Visual lexical decision tasks | None reported | None reported | 22/80 (28%) on high image words/nonwords | 99% using motor strategy |
| Letter naming | 38-58% correct during baseline | 16/26 (62% correct) | 3/26 (12% correct) | “severely impaired” |
| Stimuli/ Treatment approach | Words/letters; copied into palm using capped pen | Single letters; copied into palm using capped pen | Single letters; traced with finger | Words; traced with finger |
| Treatment outcome | Improved accuracy of letter naming, trained words and untrained words | Improved accuracy of naming letters in isolation, in strings and in words | Significant improvement in oral letter naming; improved reading of written words using LBL strategy | Improvement in reading speed using motor strategy; generalized to untreated sentence probes |

BDAE = Boston Diagnostic Aphasia Examination (Goodglass & Kaplan, 1983)

BNT = Boston Naming Test (Kaplan et al., 1983)

WAB = Western Aphasia Battery (Kertesz, 1982)

BARF = Battery of Adult Reading Function (Rothi et al., 1986)

Table 2.
TS' performance on pre-treatment assessment tasks

| | Score | % Correct | Composite Score |
|---|--------------|------------------|------------------------|
| Visual Processing | | | 98% |
| Mirror reversal letter identification (PALPA 18) | 36/36 | 100% | |
| Upper-lower case match (PALPA 19) | 26/26 | 100% | |
| Lower-upper case match (PALPA 20) | 26/26 | 100% | |
| Lexical decision (words/illegal nonwords; PALPA 24) | 56/60 | 93% | |
| Written word-picture match (PALPA 48) | 38/40 | 95% | |
| Visual Input – Verbal or Motor Output | | | 22% |
| Direct copy of words (number of letters correct) | 34/55 | 62% | |
| Case conversion: upper-to-lower | 5/26 | 19% | |
| Case conversion: lower-to-upper | 5/26 | 19% | |
| Letter naming (PALPA 22) | 12/26 | 46% | |
| Letter to sound (visually presented letter) | 3/20 | 15% | |
| Oral reading of regular words | 4/40 | 10% | |
| Oral reading of irregular words | 8/40 | 20% | |
| Oral reading of nonwords (length matched to words) | 0/20 | 0% | |
| Oral reading CVC nonwords | 4/60 | 7% | |
| Auditory Input – Verbal or Motor Output | | | 79% |
| Writing letters to dictation | 26/26 | 100% | |
| Writing regular words to dictation | 33/40 | 83% | |
| Writing irregular words to dictation | 23/40 | 58% | |
| Writing nonwords to dictation | 19/20 | 95% | |
| Recognition of orally spelled regular words | 29/40 | 73% | |
| Recognition of orally spelled irregular words | 23/40 | 58% | |
| Recognition of orally spelled nonwords | 13/20 | 65% | |
| Letter to sound (e.g., “What sound does a ‘B’ make?”) | 14/20 | 70% | |
| Sound to letter name (e.g., “What letter goes with /b/?”) | 18/20 | 90% | |
| Spelling CVC nonwords | 57/60 | 95% | |

PALPA = Psycholinguistic Assessments of Language Processing in Aphasia (Kay, Lesser & Coltheart, 1992)

Figure 1.
CT showing TS' left temporo-parieto-occipital damage

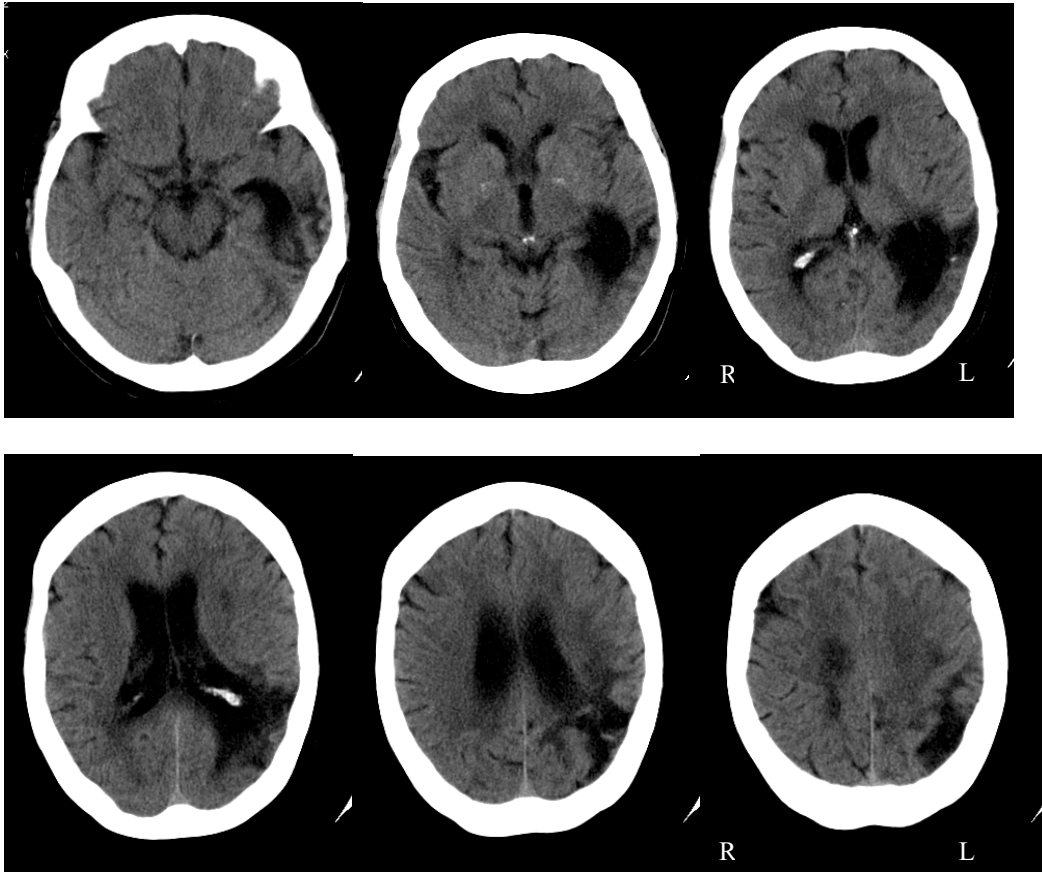


Figure 2

Sample of TS' performance on direct copy of words vs. writing words to dictation

provide
p p u vtd

greet t
g rcc

though
t o o g

gang

GANG

mile

mile

doubt

DOUBT

lose

LOOSE

grumble

GRUMBLE

drive

DRIVE

honest

honest

Figure 3
TS' letter identification performance across duration of treatment

