History

DG was a 63-year-old right-handed man who experienced a left hemisphere ischemic stroke in April 2011. Neurology notes from his hospital admission stated that he presented with new onset alexia without agraphia, and anomia. No visual field defects or other neurologic impairments were found. DG's medical history included atrial fibrillation and hypertension. An MRI shortly after his hospital admission revealed a small acute infarction with hemorrhagic conversion in the inferior left temporal lobe, involving the posterior fusiform gyrus and parahippocampal gyrus. Neurology notes at discharge interpreted the MRI findings as likely due to a venous infarction of the vein of Labbe.

DG was a warehouse supervisor but had not worked since his stroke. He had a BA in History and described himself as an avid reader. He was single and lived alone.

DG was seen in our outpatient clinic 4 months after his stroke. As part of his application for disability, a neuropsychologist evaluated him 1 month after his stroke. A summary of this evaluation stated that DG had alexia without dysgraphia, mild anomia, and mild memory impairment. When DG came to our clinic, his primary complaint was slow reading, with poor comprehension. He also acknowledged occasional word finding and memory difficulties.

Assessment Methods/Tests & Results

General Language Assessment and Cognitive Screening

DG's speech was fluent and without any noticeable word finding difficulties or syntactic/ morphological errors. The Comprehensive Aphasia Test (CAT; Swinburn, Porter, & Howard, 2005) was administered to gain an overall assessment of DG's language abilities. His mean T-score for the CAT's eight language modality scores was 61.3, just below the cut-off of 62.8 used to identify individuals with aphasia. The only modality scores to fall beneath the normal range were written comprehension (score = 50; cut-off = 58) and oral reading (score = 44; cut-off = 53). No signs of dysgraphia were noted. DG scored within the normal range on the CAT's cognitive screening subtests.

Reading Assessment

Selected subtests from the Psycholinguistic Assessment of Language Processing in Aphasia (PALPA; Kay, Lesser, & Coltheart, 1992) were administered to assess DG's reading ability. The results of these subtests are summarized below; DG's scores on the reading subtests are shown in Table 1.

Letter processing. Abstract letter identification was normal and no errors were noted on letter naming or cross-case matching tasks. Simple grapheme-to-phoneme conversion was mildly impaired. DG was unable to sound out the letters e, E, y, Y, and L, although he could name these letters.

Single word reading. DG had difficulty reading nonwords (10/24), but his real word reading was more accurate. He made few errors on real word homophone pairs (39/40) compared with nonword homophone pairs (13/20). Single real word reading was fairly accurate (77/80), with letter-by-letter reading 12/40 on low imageability words and 3/40 on high imageability words. His letter-by-letter reading approach resulted in accurate reading of these words. DG also demonstrated a mild part-of-speech effect, performing worse on functor words (14/20) than on nouns (19/20), verbs (19/20), or adjectives (18/20). On an informal task requiring DG to orally read 32 suffixed words, he scored 23/32 correct, with all errors consisting of suffix substitutions (e.g., destroyer → destroying).

No significant frequency or length effects were observed during real word reading, but there was a length effect during nonword reading. He produced an equal number of visually similar errors (e.g., plea → play) and "don't know" responses during real word reading. His nonword errors were predominantly lexicalizations (e.g., churse → church).

Semantic processing. Given the observed imageability effects during single word reading, DG's semantic access was assessed via auditory and written synonym judgment tasks. He performed normally on the auditory version but made multiple errors on the written version (47/60 correct), suggesting a modality specific interaction between semantics and reading. DG's normal performance (49/52 correct) on the three picture version of the Pyramids and Palm Trees test (Howard & Patterson, 1992) is consistent with this supposition.

Text reading. Oral reading of connected text was assessed using the Gray Oral Reading Test-D (GORT-D, Bryant & Wiederhold, 1991), Form A, passages five through nine. The results are shown in Table 2. DG's slow reading was characterized by frequent, prolonged hesitations and noticeable frustration. His pattern of oral reading errors was as follows: 39% visually related; 26% suffix substitutions; 17% functor substitutions; 8.6% "don't know"; and 8.6% semantic paralexias. DG engaged in letter-by-letter reading an average of 3 times per 100 words. Reading comprehension was significantly impaired.

Phonological processing

Because DG presented with symptoms consistent with phonological dyslexia (e.g., poor nonword versus real word reading), additional subtests from the PALPA were given to test his phonological skills. DG did well on nonword repetition (30/30), auditory rhyme judgments (58/60), and phonological segmentation of initial sounds (43/45).

Written naming and spelling

DG performed relatively well on a written picture-naming task (37/40). No spelling errors were noted on this task. Similarly, DG was able to spell 21/24 orally presented nonwords presented previously in the nonword reading subtest (PALPA 31).

Diagnostic and Prognostic Conclusions

DG was diagnosed with moderate phonological dyslexia without overt signs of dysgraphia, and a mild anomia. His poor nonword versus real word reading and difficulty reading grammatical functors and affixed words are consistent with this diagnosis.

DG's presentation was atypical in a few ways. First, his lesion in the posterior-inferior temporal lobe and fusiform gyrus was more consistent with a pure alexia, as was his tendency to engage in letter-by-letter reading. His lack of phonological impairments, given the magnitude of his reading difficulty, was unusual, as was the absence of a noticeable dysgraphia.

A literature search did not reveal any studies examining natural recovery in individuals with phonological dyslexia. Studies reporting longitudinal data on natural recovery in acquired dyslexia are few, and the results suggest that, much like aphasia, these subjects often make some recovery without treatment, that initial severity is tied to eventual outcomes, and that many individuals have persistent reading impairments (Newcombe, Hiorns, Marshall, & Adams, 1975; Wilson, 1994).

At the time of the evaluation, DG was still in the period where some natural recovery would be expected. However, at 4 months post-onset, he was already through the period of time when most natural recovery occurs after a stroke, at least for aphasia (Pedersen, Jørgensen, Nakayama, Raaschou, & Olsen, 1995). Additionally, he had not noticed any reading improvement since his stroke. Given these points, significant improvement in reading, via natural recovery, was not expected.

The positive prognostic indicators for improved reading after treatment were DG's high degree of motivation, no history of developmental reading impairments, no significant language impairments outside of reading, and no visual-perceptual impairments. The negative prognostic signs were his reported mild memory impairment, lack of social support, and his apparent high degree of frustration and anxiety when reading. Overall, his prognosis was fair to good for improved reading.

Management Recommendations and Procedures

The goals of treatment, and treatment approach, were chosen in collaboration with DG. DG wanted to improve the speed and comprehension of his reading, and to practice reading stories from online news sources.

Given my review of the literature in the course of writing this paper, and my current hypothesis about why DG presented with his unique constellation of symptoms, a review of the treatment literature on pure alexia, in addition to phonological dyslexia, would be appropriate in this section. However, when I was in the process of deciding on a treatment, my focus was on reviewing the phonological dyslexia treatment literature. For this reason, I will only discuss the literature I reviewed when preparing for this client.

Two general approaches have been used in the rehabilitation of individuals with phonological dyslexia: phonological treatments, usually focusing on grapheme-to-phoneme conversion or bigraph-to-biphone conversion; and Multiple Oral Rereading (MOR; Moyer, 1979). Both of these approaches have been shown to improve reading, although the results are mixed.

The major weakness of the phonological approach, with respect to DG's goals, is the paucity of outcome measures showing improvement beyond the single word level. Most of the reviewed studies did not report outcome measures for reading rate or text comprehension (Beeson, Rising, Kim, & Rapcsak, 2010; Bowes & Martin, 2007; Friedman & Lott, 2002; Kendall, McNelil, & Small, 1998; Kendall, Conway, Rosenbek, & Gonzalez Rothi, 2003; Mitchum & Berndt, 1991; Yampolsky & Waters, 2002).

There are a few phonologically based treatment studies that reported text reading comprehension and rate outcomes, but the effectiveness of these treatments are not convincing. Minimal to mild improvements in text reading comprehension (Conway, Heilman, & Rothi, 1998; Kim & Beaudoin Parsons, 2007) and reading rate (Conway et al., 1998) have been reported. The most robust improvements were demonstrated by Peach's (2002) subject DB, who improved his reading comprehension of untreated passages from 62% to 84% accuracy, and reading rate from 67 to 94 words per minute (wpm). However, DB was 3.5 months post-stroke at the end of treatment, so these gains may have been due to natural recovery.

MOR appeared to be a better fit for DG, since it satisfied his desire to practice reading material he enjoyed and home practice is straightforward. This was important since DG lived alone and could not rely on others for assistance. Finally, the treatment literature reported outcomes that were closely aligned with DG's goals and showed comparable, if not greater, effectiveness.

MOR was designed to increase the speed of reading at the text level. This treatment requires clients to read a single passage of text multiple times per day. Since Moyer's (1979) original study, a small collection of case and case-series studies with varying degrees of experimental control (Cherney, 2004; Kim & Russo, 2010; Lacey, Lott, Snider, Sperling, & Friedman, 2010) and single subject designs (Beeson, 1998; Moody, 1988) have reported improvements in reading by subjects across a continuum of severity, from relatively mild phonological text dyslexia to deep dyslexia. Out of the eight total subjects in these studies, seven improved their reading rate. The duration of treatment ranged from 8 weeks (Lacey et al., 2010) to 10 months (Beeson, 1998), with an increase in reading rate from 17% (Lacey et al., 2010) to 112% (Beeson, 1998).

There is less information about how MOR affected these subjects' oral reading accuracy and comprehension of untreated texts. Of the studies that reported these data, subjects improved oral reading accuracy but did not significantly improve reading comprehension (Kim & Russo, 2010; Lacey et al., 2010). The lack of improvements in reading comprehension may have been due to the kind and degree of impairments present in these subjects. Kim and Russo's (2010) subject had a nonfluent aphasia, apraxia of speech, and deep dyslexia. His reading rate improved 33% to 6.27 wpm; however, these changes may not have been large enough to support better comprehension. The subjects in the Lacey et al (2010) study, on the other hand, began treatment with relatively good reading comprehension, so there was less room for improvement.

Kim and Russo (2010), after a review of the treatment literature, outlined five criteria for a successful response to MOR: 1) good single word reading and comprehension; 2) preserved letter identification and naming; 3) fairly good oral single word reading and comprehension; 4) no significant impairments of verbal expression; and 5) no cognitive impairments, particularly attention. Aside from DG's reported mild memory impairment, he met all of the criteria and was considered a good treatment candidate for MOR.

In summary, it appears that MOR is an effective treatment for most subjects with phonological dyslexia. At the beginning of treatment, I hypothesized that improving DG's reading rate would improve comprehension by reducing the burden on his memory resources. Any gains in reading accuracy would also support better comprehension through more accurate semantic and syntactic processing of text.

Procedures

DG was seen for therapy once a week, for 45 minutes per session. Treatment passages were 300-500 words in length and came from online news sources. DG was given a new passage to read each week. Each session began with DG reading the previous week's passage without assistance. Reading rate and number of oral reading errors were calculated from these practiced passages. After this initial reading, DG was given a new passage to read without assistance and the same measurements were taken. DG then read this new passage a second or third time, with the clinician assisting him with reading difficult words. A printed copy was given to DG to practice reading at home 30 minutes a day. DG was given a log to record his home practice.

Data Documenting Outcome of Treatment

Single Word Reading

Only those PALPA subtests that DG had difficulty with during the initial testing were re-administered. Given the PALPA's lack of standardization and the relatively small number of stimuli, particularly when broken down by psycholinguistic attribute (e.g., imageability), the following results should be interpreted with caution.

DG improved his letter-to-sound production, with no errors on this task; nonword reading improved by 37.5%. DG made a 31.6% improvement on the written synonym judgment task, with better performance on low imageability words.

Text Reading

Generalization to untreated text was assessed by the GORT-D. At the beginning of treatment, DG was given the GORT-D, Form A. Form B was given after 6 weeks of treatment, and form A was re-administered at the end of treatment. By the end of treatment, reading rate improved by 23% and the number of oral reading errors was reduced by 74%; reading comprehension improved by 70% (see Table 2).

Weekly probes using new practice passages were used to measure changes in reading rate and accuracy over the course of treatment. Baseline measures were taken over 3 weekly sessions. The third baseline measurement came from the initial reading of DG's first practice passage.

After a week of practice, DG read his practiced passages faster and more accurately (avg. wpm = 56.3; avg. oral reading errors per 100 words = 4.5). Figure 1 shows the results of the baseline and treatment probes (data from session 8 were lost). Results from the GORT-D were graphed along with these measures for comparison (Fig. 1). Consistent with the results of the GORT-D, a visual analysis of the treatment probes suggests a gradual improvement in oral reading accuracy, and possibly reading rate. However, not enough baseline measures were taken to establish a stable baseline and to confidently state that any observed improvements were due to treatment and not natural recovery. DG's more accurate reading of GORT-D passages versus the practice passages was likely due to the lower grade level of the GORT-D passages.

Rationale for Termination of Treatment and Follow-up Recommendations

Treatment ended after 4 months. I was leaving to work at another medical center and DG did not want to continue with another therapist, possibly because he was frustrated by his lack of meaningful improvement. I noticed DG growing more frustrated in the second half of treatment and reducing the amount of home practice he completed. By the end of the treatment phase, DG was practicing approximately 1 hour a week.

In the second half of my 4 months working with DG, I made adjustments to the treatment procedures in an attempt to increase his engagement. My goals were to increase his experience of success during his home practice and increase the personal relevancy of the reading passages. At the end of each session, DG was given an audio file consisting of an oral reading of the week's practice passage. DG was instructed to read along with the audio recording before reading the passage on his own. To increase the relevancy of the news passages, and DG's control over the treatment process, he was encouraged to bring in news stories that he wanted to read.

In the end, none of the changes described above improved his treatment adherence. DG was not interested in other treatment approaches, such as word- or grapheme-level treatments. I recommended that DG consider returning to therapy sometime in the future. DG did return to our clinic; however, my understanding is that he discharged himself from treatment a month after re-starting.

Integration of Content with Literature

Diagnosis

There are two prevailing hypotheses that attempt to explain the lexicality effect in phonological dyslexia. Dual-route model proponents believe impaired grapheme-to-phoneme conversion (GPC) is the cause (Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001) while connectionist model proponents (Crisp & Ralph, 2006) believe the cause is a general phonological impairment. Evidence exists for both the GPC (Tree & Kay, 2006; Vliet & Miozzo, 2004) and the phonological account (Crisp & Ralph, 2006; Rapcsak et al., 2009).

DG's lack of dysgraphia and good performance on phonological tasks argue against the presence of a general phonological impairment. On initial testing, he had mild difficulty converting letters to sounds, which is consistent with GPC impairment. However, neither phonological processing nor GPC correspond to DG's lesion site. These skills are believed to reside in left hemisphere perisylvian cortex (Jobard, Crivello, & Tzourio-Mazoyer, 2003; Price, 2012; Rapcsak et al., 2009).

DG's lesion in the posterior fusiform gyrus and adjacent temporal lobe, and his occasional letter-by-letter reading, is more consistent with pure alexia (Cohen et al., 2004; Mani et al., 2008). A literature search to find cases that resembled DG (i.e., similar symptoms and lesion profile) was conducted. Two cases were found that were roughly similar to DG (Buxbaum & Coslett, 1996; Friedman et al., 1993). Both subjects had features of both pure and phonological dyslexia and had ventral occipitotemporal lesions.

Of these cases, Buxbaum and Coslett's (1996) subject, JH, is the closer match to DG. JH was described as having features of both pure alexia and partially recovered deep dyslexia. He had a lesion in the left inferior temporal-occipital cortex, with involvement of the parahippocampal gyrus, and a lesion in the left lateral geniculate. He did not have aphasia or dysgraphia.

Like DG, JH presented with poor nonword compared with real word reading, and imageability, morphological, and part-of-speech effects. He also tended to lexicalize nonwords. Real word errors were predominantly visual. JH also engaged in letter-by-letter reading.

One account, given by Buxbaum and Coslett (1996), for JH's pattern of reading impairments, was that weakened pre-lexical visual analysis resulted in weak activation of intact orthographic entries. At the semantic level, this weak lexical activation may have been adequate for semantically rich words, but not for grammatical functors and other low imageability words. By this account, nonwords would also be difficult since they have no semantic representations.

Other studies have shown that lexical variables other than word length can affect reading in pure alexia. Imageability (Behrmann, Plaut, & Nelson, 1998; Behrmann, Shomstein, Black, & Barton, 2001), concreteness (Behrmann et al., 1998), and age of acquisition (Cushman & Johnson, 2011) effects have been reported. These effects have been described in the context of an interactive activation model of reading (Behrmann et al., 1998; 2001; Cushman & Johnson,

2011) where top-down (e.g., phonology and semantics) and bottom-up visual input interact in the process of reading. Price and Devlin (2011) hypothesized that the ventral occipitotemporal cortex was an interface between higher-order phonological and semantic associations and visual input.

In summary, DG fit the syndrome of phonological dyslexia but his letter-by-letter reading and lesion location suggested a pure alexia. It is possible that he did not have phonological or GPC impairments. Rather, pre-lexical processing was damaged, resulting in more difficulty reading words that provided less top-down support.

Treatment

Without a control task and stable baselines, it is difficult to say whether DG's improvements were due to treatment or natural recovery. His improvement on single word reading tasks from the PALPA may have been a sign of natural recovery, since he would not have been able to use the top-down contextual strategies learned during MOR treatment to decode these words (Tuomainen & Laine, 1991). However, evidence suggests that MOR also improves the reading of untreated single words, both in individuals with phonological dyslexia (Beeson, 1998; Kim & Russo, 2010; Lacey et al., 2010) and pure alexia (Beeson, 1998). Given this evidence, it is difficult to say whether DG's improvements were due to natural recovery or not.

A Brief Self-Critique

I believe that MOR was a good fit for DG and that his lack of home practice, in the second half of treatment, reduced the gains he could have made. My main failure, as I see it, was that I did not adequately respond to DG's changing pattern of home practice. Motivation is a dynamic state that can be influenced by many variables. Important determiners of motivation are accurate deficit awareness, satisfaction with treatment, outcome expectancy, and self-efficacy (Drieschner, Lammers, & van der Staak, 2004; Lequerica & Kortte, 2010). Periodic self-assessment measures probing these variables may have allowed me to identify issues that could have been addressed early. Additionally, clinician-based ratings of engagement in therapy, such as the Pittsburgh Rehabilitation Participation Scale (Lenz et al., 2004), may have contributed to early detection of changes in DG's attitude towards treatment.

Not long after I finished treatment with DG, I received training in Motivational Interviewing (MI). MI is "a client-centred, directive method for enhancing intrinsic motivation to change by exploring and resolving ambivalence" (Miller & Rollnick, 2002, p.25). Based on my current experience using MI, I recognize that I was too prescriptive with DG and spent too much time trying to convince him to continue his home practice, rather than engaging him in conversations that would have allowed him to voice his concerns and frustrations. By not exploring issues in a truly collaborative way, I allowed my therapeutic relationship with DG to gradually decline. This decline appeared to coincide with my report to him that I was taking a new job and may not be able to complete treatment with him. Whether this announcement played a negative role on DG's motivation is hard to say. If it was an issue, then more open lines of communication would have afforded us opportunities to discuss the matter and prepare for a transition to another therapist.

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Table 1

Reading Subtests from the PALPA Pre- and Post-treatment

PALPA subtest		Pre-treatment	Post-treatment	Subtest total % change
19 - Upper Case - Lower Case Letter Matching		26/26	NT	
22 - Letter Naming/Sounding	Naming	52/52	NT	15.5%
	Sounding	45/52	52/52	
25 - Imageability & Frequency Visual Lexical Decision	HI + HF	15/15	15/15	
	HI + LF	14/15	15/15	
	LI + HF	13/15	15/15	
	LI + LF	12/15	12/15	5.6%
	Nonword	60/60	60/60	
28 - Homophone	Regular	20/20	19/20	
	Irregular	19/20	20/20	3.8%
	Nonwords	13/20	15/20	
29 - Letter Length Reading		23/24	NT	
31 - Imageability & Frequency Reading	Overall	77/80	NT	
	High Frequency	40/40	NT	
	Low Frequency	39/40	NT	
	High Imageability	38/40	NT	
	Low Imageability	37/40	NT	
32 - Grammatical Class Reading	Noun	19/20	20/20	
	Verb	19/20	20/20	
	Adjective	18/20	19/20	7.1%
	Functor	14/20	16/20	

PALPA subtest	Pre-treatment		Post-treatment	Subtest total % change
36 - Nonword Reading vs. Naming to Oral Spelling	3 - Letter	4/6 - 6/6	5/6 - NT	
	4 - Letter	2/6 - 6/6	4/6 - NT	
	5 - Letter	1/6 - 5/6	1/6 - NT	37.5%
	6 - Letter	1/6 - 4/6	1/6 - NT	

Table 1 cont.

PALPA subtest	Pre-treatment		Post-treatment	Subtest total % change
49 - Auditory Synonym Judgment		58/60	NT	
50 - Written Synonym Judgments	High Imageability	28/30	28/30	
	Low Imageability	19/30	25/30	31.6%

Table 2

Gray Oral Reading Test-D Pre- and Post-treatment

Phase	wpm	Errors per 100 words	% Comprehension
Pre-treatment	24.3	4	40
Mid-treatment	33.1	1.9	60
Post-treatment	29.9	1.04	68

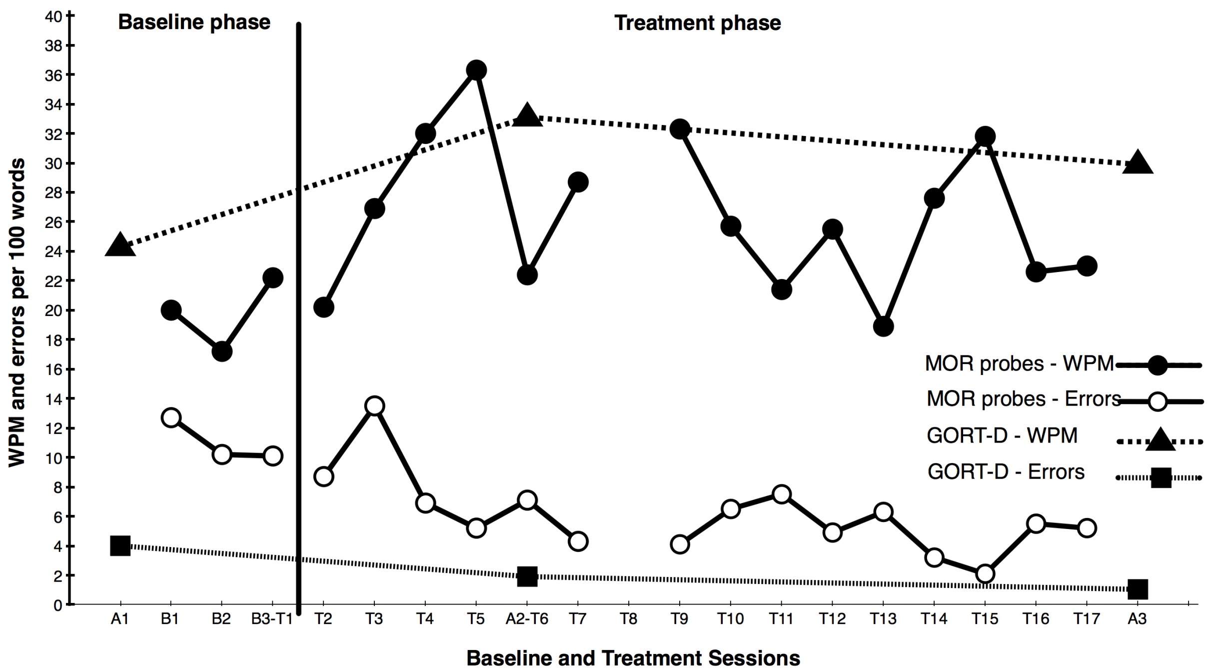

Figure 1. Multiple oral rereading baseline (B) and treatment (T) probes. GORT -D assessments (A).