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Comparing uni-modal and multi-modal therapies for improving writing in acquired dysgraphia after stroke

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For submission to: Neuropsychological Rehabilitation

Keywords: therapy, aphasia, spelling, writing, multi-modal

Short title: Multi-modal and uni-modal dysgraphia therapies

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Acknowledgements:

This work is supported by an Economic and Social Research Council bursary to Lindsey Thiel [Award number ES/I020233/1]
Abstract

Background: Writing therapy studies have been predominantly uni-modal in nature; i.e. their central therapy task has typically been either writing to dictation or copying and recalling words. There has not yet been a study that has compared the effects of a uni-modal to a multi-modal writing therapy in terms of improvements to spelling accuracy.

Aims: A multiple-case study with eight participants aimed to compare the effects of a uni-modal and a multi-modal therapy on the spelling accuracy of treated and untreated target words at immediate and follow-up assessment points.

Methods and Procedures: A cross-over design was used and within each therapy a matched set of words was targeted. These words and a matched control set were assessed before as well as immediately after each therapy and six weeks following therapy.

Outcomes and Results: The two approaches did not differ in their effects on spelling accuracy of treated or untreated items or degree of maintenance. All participants made significant improvements on treated and control items; however not all improvements were maintained at follow-up.

Conclusions: The findings suggested that multi-modal therapy did not have an advantage over uni-modal therapy for the participants in this study. Performance differences were instead driven by participant variables.
Introduction

A substantial body of research has investigated the effects of deficit-focused writing therapies for people with aphasia (e.g. Beeson, 1999; Luzzatti, Colombo, Frustaci, & Vitolo, 2000; Rapp, 2005; Raymer, Strobel, Prokup, Thomason, & Reff, 2010; Schmalzl & Nickels, 2006). These therapies have been shown to be successful in improving single-word writing in people with a range of types and severities of dysgraphia. One factor that has not yet been investigated is the effect of multi-modality within writing therapy tasks.

The concept of people with aphasia relearning a target word through completing tasks in different modalities, i.e. through saying the word, gesturing, writing the word and making semantic, phonological or orthographic decisions about the word, is certainly not novel. Howard, Patterson, Franklin, Orchard-lisle and Morton (1985) advocated a multi-modal approach to eliciting words from patients within a spoken naming therapy. They investigated the effects of semantic and phonological cues as prompts in picture naming and showed through different experiments that naming could be improved by asking participants to carry out spoken word to picture matching tasks, as well as written word to picture matching, semantic judgement, repetition and rhyme judgement tasks and through being shown a picture together with a spoken word that rhymes with the target. More recently, a study by Weill-Chounlamountry, Capelle, Tessier & Pradat-Diehl (2013) investigated the effects of a computer-delivered phonological multi-modal therapy for naming. The participant with fluent aphasia was presented with a picture of an object and then completed a sequence of tasks including rearranging the letters, verbally repeating the syllables, graphemes and whole word, coping letters, syllables and the whole word, delayed copying, writing the name and then finally saying the word. This therapy led to significant improvements to oral naming of trained and untrained items, which was maintained at 3 month follow up.

Rose and colleagues (e.g. Attard, Rose & Lanyon, 2013; Rose & Douglas, 2008; Rose, Douglas & Matyas, 2002; Rose, Attard, Mok, Lanyon & Foster, 2013) have investigated the efficacy of combining verbal and gesture tasks to improve naming. Rose & Douglas (2008) and Rose et al. (2002) found this combined approach to be equally as effective as both verbal and gesture therapies for participants with both lexical-semantic and phonological naming impairments. Recently, Rose et al. (2013) compared constraint-induced therapy (CIATplus) to a multi-modal treatment (M-MAT, Rose & Attard, 2011) for their effects on naming
accuracy in 11 participants with aphasia. M-MAT employed a cueing hierarchy, in which participants were asked to gesture, draw, copy and repeat the target words. CIATplus consisted of a cueing hierarchy of phonemic and written cues, with participants only being asked to name the item. It was found that both treatment approaches were equally efficacious in terms of mean effect size across participants for noun and verb naming, although 6 participants expressed a preference for M-MAT, whereas only 3 preferred CIATplus.

Some writing therapy studies have also demonstrated successful outcomes following therapy approaches that have been multi-modal (Ball, de Riesthal, Breeding, & Mendoza, 2011; Beeson and Egnor, 2006; Behrmann, 1987; Cardell & Chenery, 1999; Carlomagno, Iavarone, & Colombo, 1994; de Partz, Seron, & Vanderlinden, 1992; Hatfield & Weddell, 1976; Schmalzl & Nickels, 2006; Weekes & Coltheart, 1996). For example, Ball et al. (2011) modified Anagram and Copy Treatment (ACT) and Copy and Recall Treatment (CART) for three participants with aphasia by incorporating naming and spoken repetition. Within sessions, participants were first asked to name a drawing. If they could not do this, they were asked to repeat the word spoken by the therapist three times. They then continued with ACT, which involved writing the picture name, arranging letters of the word into the correct order, copying the written word and then writing the word from memory. At home, participants were encouraged to repeat target words that they heard in video clips and then to proceed with CART (copying words and then writing them from memory). All participants improved their written naming accuracy of treated items and one participant showed generalisation to untreated words.

There has been one published study which has compared uni-modal and multi-modal writing therapies. Schwartz, Nemeroff and Reiss (1974) compared an “experimental” writing therapy given to eight people with aphasia to a “control” condition of multi-modal therapy provided to six people with aphasia who were matched on age, months post brain injury, education and pre-therapy scores on the Porch Index of Communicative Ability (PICA; Porch, 1971). In the experimental condition, participants completed a range of writing tasks for each item, including writing the alphabet from memory, written picture naming, writing to dictation after hearing the word once or three times and, finally, writing words that had been placed into a spoken sentence. The multi-modal therapy incorporated the following tasks: spoken picture naming, spoken word-to-picture matching, reading aloud, written picture naming and
repetition. The same words were targeted in both conditions. However, within each condition, sets of words were split between different tasks. For example in the multi-modal condition, some words were trained with spoken picture naming while others were trained with reading aloud. Therefore, in the multi-modal therapy individual words were not targeted in different modalities. Success in therapy was measured using the PICA. No significant difference was found between the two groups’ scores. However, the experimental group made more improvement compared to baseline than the control group.

These studies provide initial evidence that a multi-modal therapy can be effective for improving writing in people with dysgraphia; however, they do not indicate whether a multi-modal approach is more effective than a uni-modal approach for improving writing, i.e. leads to greater accuracy scores across matched sets. Multi-modal treatments are often viewed as being more effective than uni-modal treatments by speech and language therapists, although there is a lack of evidence to support this claim (Rose & Douglas, 2008). Lexical writing therapies that have been uni-modal in nature in which participants copy and recall words or write words from dictation with cues (e.g. Beeson, 1999; Schmalzl & Nickels, 2006) have been shown to be successful in terms of gains to treated items; however these gains have often not been maintained and have seldom led to generalisation to untreated words (exceptions have usually been to participants with graphemic buffer disorder, e.g. Mortley et al., 2001 Panton & Marshall, 2008; Pound, 1996; Rapp, 2005; Rapp & Kane; 2002; Raymer, Cudworth & Haley, 2003; Sage & Ellis, 2006; Thiel & Conroy, 2014).

Connectionist theories of language processing, such as the Primary Systems Hypothesis (Patterson & Lambon, 1999) and the Triangle model (Plaut, McClelland, Seidenberg & Patterson, 1996, see Figure 1) have conceptualised specific skills such as reading and writing as being underpinned by an interaction between the three core underlying systems: semantics, phonology and orthography. Therefore, disruption to any of these core systems due to brain damage will result in a disruption to reading and writing. Despite a rich and complex literature, there have been relatively few studies which have applied connectionist principles to neurorehabilitation. The few available language-focused studies have tended to address anomia (e.g. Abel, Willmes & Huber, 2007; Abel, Huber & Dell, 2009) and compared different connectionist models in terms of their utility for treating symptoms and predicting therapy gains. Similar to work within connectionist modelling of reading and dyslexia, studies on dysgraphia have modelled spelling acquisition and breakdown in simulations of
brain damage (e.g. Loosemore, Brown & Watson, 1991) but not yielded treatment principles and investigations.

At least three hypotheses can be proposed to support the contention that multi-modal therapy may be distinct from and potentially more beneficial than uni-modal in terms of variables such as extent of accuracy achieved, likelihood of generalisation, or maintenance of therapy gains. Firstly, it is hypothesised, that consistent with connectionist models of language processing and distributed representations (e.g. Welbourne & Lambon Ralph, 2007), in a multi-modal therapy, distributed semantic, phonological and orthographic representations will be activated for each target word, which will strengthen connections and weight adaptations between language-related units (semantics, phonology, orthography). This may lead to more interactive and robust processing, and therefore more accurate and lasting learning of written words relative to a uni-modal therapy in which words have just been copied and recalled. Secondly, as a consequence, however, because each word will be copied less frequently, multi-modal processing may well have the potential disadvantage of showing a slower trajectory of increasing accuracy scores relative to uni-modal therapy, i.e. fewer items may be ‘relearnt’ over set time frames. Finally, targeting phonology, semantics and orthography will strengthen these underlying systems, which may result in improved writing accuracy for untreated words, i.e. greater evidence of generalisation effects following multi- as opposed to uni-modal therapy.

[Insert Figure 1 about here]

The aim of this within-participants multiple case study was to answer the following questions:

1. Is a multi-modal therapy more effective than a uni-modal therapy in improving spelling accuracy across matched sets?
2. Does a multi-modal therapy lead to a greater degree of generalisation to untreated words than a uni-modal therapy?
3. Is a multi-modal therapy more effective than a uni-modal therapy in terms of maintenance of learning effects across matched sets once treatment has concluded?
Method

Recruitment

Eight participants were recruited to this study. To be included participants had to have an acquired spelling impairment following a stroke. They had to be at the chronic stage of their brain injury (i.e. post six months). They had to have sufficient visual acuity and motor ability for handwriting. Finally they needed to be monolingual speakers of English. Potential participants were excluded if they had a severe impairment in reading or auditory comprehension (i.e., in the lower 50% of the aphasic population). These skills were assessed using subtests from the Comprehensive Aphasia Test (Swinburn, Porter & Howard, 2004).

Participants

Background Assessments

The participants completed a battery of linguistic and writing assessments. Tables 1, 2 and 3 display participants’ demographic information, screen scores and assessment results on spelling and language assessments. Participants have been ordered according to total baseline spelling scores on the PALPA word spelling subtests, with the most impaired to the left and the least impaired to the right. These tables are followed by a description of each participant’s language and writing skills. All assessments were administered by the first author.

[Insert Tables 1, 2 and 3]

Description of participant’s linguistic and writing skills

JP suffered a single left hemisphere stroke in 2004 subsequent to surgical removal of a brain tumour in 1999. She presented with unimpaired spoken language within conversation, although her scores on the BDAE revealed impairments across all language skills. She scored 36/52 on the Pyramids and Palm Trees Test (matching pictures; Howard & Patterson, 1992),
which indicated impaired semantics. When writing words to dictation, she converted sounds to letters aloud (a strategy she had learnt in previous therapy). She wrote 9/24 non-words to dictation and showed a significant length effect, when 3 and 4 letter words on the PALPA 39 were compared statistically to 5 and 6 letter words (p < .001, Fisher's exact test) and a significant imageability effect (p = .02, Fisher's exact test). She demonstrated a marked disparity in her ability to write regular and irregular words (although the difference was not statistically significant). Furthermore, she often regularised irregular words, resulting in errors such as ‘serkle’ for circle, ‘clok’ for clock, ‘speck’ for speak, ‘elefant’ for elephant, and ‘lern’ for learn. Her difficulty with irregular words, her tendency to rely on phoneme to grapheme conversion rules as opposed to stored representations and her resulting regularisation errors suggested that she had surface dysgraphia, a central (linguistic) dysgraphia syndrome, in which individuals have more difficulties spelling irregular words than regular words and make regularisation errors (e.g. laugh may be spelt as ‘larf’) (Rapcsak, Henry, Teague, Carnahan, & Beeson, 2007). Relative to the other participants she had a low score (18/27) on the copying task on the CAT (Swinburn, Porter & Howard, 2004), also suggesting damage to the peripheral components of writing, i.e. accessing the appropriate allographs (letter shapes) or to the motor programmes responsible for letters being written or typed (Beeson & Rapscak, 2002).

DM had non-fluent aphasia following a single left hemisphere stroke in 2007. He communicated effectively with spoken language, however, predominantly with nouns due to agrammatism. He did not show any effects of length, frequency or regularity. However, he did show a significant imageability effect on the PALPA 40 (Kay, Lesser & Coltheart, 1992) (p = .03, Fisher's exact test). He was unable to write any non-words to dictation. He made occasional semantic errors, for example, ‘dish’ for spoon and ‘post’ for letter as well as letter addition, omission, substitution and movement errors, for example ‘stemp’ for stamp and ‘dace’ for dance. Some of his responses were unrelated to the target with less than 50% letters correct, e.g. ‘rillir’ for rabbit and ‘hidder’ for think. He had more difficulty writing verbs than nouns, and in many cases could not retrieve any of the word. His writing impairment could best be described as deep dysgraphia due to his inability to write non-words, his semantic errors and his imageability effect. The term deep dysgraphia has been used to describe a central (linguistic) dysgraphia syndrome which includes symptoms such as the production of semantic errors such as ‘fork’ for knife, impaired non-word spelling, and
KR presented with severe non-fluent aphasia caused by a left hemisphere stroke in 2008. She communicated by producing a few single spoken words, writing single words and short sentences, and drawing. On the PALPA 40 (Imageability and Frequency Spelling) she scored significantly lower on low imageability words than high imageability words (p < .001, Fisher's exact test) and on the PALPA 39 she showed a length effect (p = .03, Fisher's exact test). KR’s errors on these assessments included semantic errors (e.g. ‘hand’ for glove), phonological errors (e.g. ‘knot’ for knock) and letter addition errors (e.g. ‘yachet’ for yacht), with the latter being the most common error type. She did not write any non-words correctly on the PALPA 45. Based on her difficulty in spelling non-words, her imageability effects and her errors, KR has been classified as having deep dysgraphia (Whitworth et al., 2005). Furthermore her length effect and errors are characteristic of graphemic buffer disorder (Miceli, Silveri & Caramazza, 1985). In contrast to central dysgraphias (surface, phonological and deep) which are caused by underlying linguistic deficits (Ellis & Young, 1988), graphemic buffer disorder is a “peripheral dysgraphia” (Lesser & Milroy, 1993) that has been described as being caused by a deficit in the short-term storage mechanism for the orthographic representations of words while writing is planned and executed. Symptoms include length effects and the following error types: letter additions (tractor → TRACCTOR), substitutions (tractor → TRAPTOR), omissions (tractor → TRACOR) and transpositions (tractor → TRATCOR) (Rapp, 2005; Sage & Ellis, 2006). Furthermore, words and non-words are usually affected similarly (Sage & Ellis, 2006); however, KR had more difficulty writing non-words with most responses being completely unrelated to the target (‘joie’ for bem; ‘kawhs’ for nar).

AD had severely impaired expressive language due aphasia and apraxia of speech following a left hemisphere stroke in 2009. Her speech was fluent but with frequent phonological errors. She did not demonstrate any effects of imageability, frequency, regularity or word length on the PALPA subtests. Her errors on these word and non-word spelling assessments included letter additions (e.g. ‘ghoste’ for ghost), omissions (e.g. ‘ream’ for realm and ‘hoch’ for hoach), transpositions (e.g. ‘sntie’ for snite) and substitutions (e.g. ‘rorrin’ for robin). She correctly spelled 10 non-words to dictation, indicating that she had some ability to convert phonemes to graphemes. Her symptoms do not point clearly towards any one dysgraphic
syndrome. However, her errors and the fact that her words and non-words were similarly affected (41.7% correct non-words; 53.8% correct words) suggest that she may have had a graphemic buffer disorder (Rapp, 2005; Sage & Ellis, 2006), although she did not show an effect of length.

Since suffering a left hemisphere stroke in 1995, JB presented with aphasia, but also severe dysarthria and apraxia of speech. Her writing, which she had learnt to do with her non-dominant left hand, was very slow, effortful and often quite unintelligible. On the PALPA subtests, she did not show effects of imageability, frequency or regularity. She only managed to write two non-words to dictation and sometimes lexicalised them (e.g. ‘fond’ for fon and ‘pearl’ for birl). Her incorrect responses were either no responses, included less than 50% of the letters in the target word (e.g. ‘s’ for strength; ‘utable’ for choose), or were letter addition or omission errors (e.g. ‘texte’ for text; ‘stberry’ for strawberry). Her impaired non-word writing and her unrelated responses were characteristic of phonological dysgraphia, a central (linguistic) dysgraphia sub-type that describes people with impaired non-word spelling, lexicality effects (where a non-word such as SOAF is spelt as a phonologically similar stored word such as SOAP) (Rapcsak, Beeson, Henry, Leyden, Kim, Rising, Andersen & Cho, 2009) and imageability effects (Whitworth, Webster & Howard, 2005).

SR had a left hemisphere stroke in 2007 and then another in 2010. His language skills appeared to be intact within conversations; however background language assessments revealed impaired naming, auditory comprehension and semantic access. He also had residual writing difficulties. On the PALPA subtests, he did not show effects of length, imageability or frequency. However, he did have more difficulty with spelling exception words than regular words on the PALPA 44 (p < .001, Fisher's exact test). Furthermore, he was able to spell 19/24 non-words correctly. The majority of his errors were regularisations of exception words (generally the low frequency ones). For example, he wrote ‘sigaret’ for cigarette, ‘nefew’ for nephew, ‘nolidge’ for knowledge and ‘perswade’ for persuade. Based on these assessment results, SR’s spelling impairment can be described as surface dysgraphia (Rapcsak et al., 2007).

MB had a single left hemisphere stroke in 2010, which resulted in fluent aphasia with occasional word-finding difficulties. He did not display effects of imageability, frequency, regularity or word length. His errors on these tests were a mixture of letter omission errors (e.g. ‘churh’ for church) and no responses. He did not spell any non-words to dictation.
correctly and on ten occasions showed lexicality effects (e.g. ‘hug’ for cug, ‘fog’ for fon). These assessments suggest that his predominant difficulty was with converting phonemes to graphemes with the absence of a stored representation of the word. He therefore fitted the profile of phonological dysgraphia (Rapcsak et al., 2007).

Following a single, left hemisphere stroke in 2010, EB had fluent speech with occasional phonological errors and word finding difficulties. She did not show effects of length, frequency or regularity. However, she did show an imageability effect on the PALPA 40 ($p = .02$, Fisher's exact test). She only wrote four non-words correctly to dictation, indicating a more severe impairment spelling non-words compared to words. Her responses often consisted of correct initial and final spellings with the middle of the word being incorrect. This was especially true for longer words that could be segmented into morphemes. For example, she spelt impairment as ‘impartment’, television as ‘television’ connection as ‘conation’ and accommodation as ‘accondation.’ Many of her incorrect responses were letter omission errors (e.g. ‘gradfather’ for grandfather and ‘lanuage’ for language). However, she also frequently added grammatical morphemes onto dictated words (e.g. ‘enjoyed’ for enjoy and ‘strawberry’s’ for strawberry). The difficulties with converting phonemes to graphemes within non-words and the imageability effect suggest that EB had phonological dysgraphia (Rapcsak et al., 2007).

In summary, the participants had a broad range of dysgraphia severities and types, with surface, phonological and deep dysgraphias being represented and two participants showing possible symptoms of graphemic buffer disorder. Some had mixed types of dysgraphia, with symptoms of more than one syndrome. It is important to note that many people who present with dysgraphic symptoms do not fit neatly into any one category. According to Beeson and Rapcsak (2002) the subcategories of dysgraphia can be useful for communicating clusters of symptoms, but are best supplemented with descriptions of impaired and preserved processes.

**Therapy**

Baseline Spelling Assessment

With the assistance of the first author and family members, participants generated a list of functionally useful words for therapy. Additionally, participants were assessed on word lists
generated by the first author. These consisted of words from several spelling, reading and picture naming assessments, such as the Object and Action Naming Battery (OANB, Druks & Masterson, 2000), the Boston Naming Test (Kaplan, Goodglass & Weintraub, 2001) and the Baxter and Warrington Spelling Test (1994), as well as additional words considered to be useful for email writing by the first author (e.g. meeting, appointment, holiday, stroke). Participants were asked to spell the word lists as well as the self-chosen items to dictation on three occasions. Responses were considered correct and were given a score of 1 if each letter was in the correct place. Incorrectly spelt words were scored as 0. A 20 second cut-off was given for participants to respond to each word. 120 words that were spelt incorrectly on two or three occasions were selected for three word lists for each participant which were divided in the following way: two lists were used for the two therapy manipulations and one list was not treated at all (control condition). These sets were matched for word length (phonemes and letters), word frequency, imageability, regularity and word class (i.e. number of nouns, verbs and adjectives).

Procedure

Two different therapies were provided to each participant: multi-modal therapy and uni-modal therapy. In order to control for order of therapy effects, these therapies were provided within a cross-over design (see Figure 2). Half of the eight study participants (Group 1) had uni-modal therapy and then multi-modal therapy, and the remaining participants (Group 2) had the therapies in reverse order. Participants received 5 hourly sessions of each therapy (10 hours in total) which took place over three weeks with a two week break between the two types of therapy.

[Figure 2 about here]

Uni-Modal Therapy

A schematic representation of uni-modal therapy can be seen in Figure 3. First, the participant was asked to copy the written target word from a card. The first author then gave feedback on the accuracy of the response (correct or incorrect). If it was copied incorrectly or
no response was given within 20 seconds, the therapist asked the participant to copy the word from the card two more times. The second time, she commented on its accuracy. If the word was copied correctly on the first attempt, the target word and the participant’s response were covered and the participant was asked to write it from memory. Feedback was then given by the therapist on whether the production was accurate or inaccurate. If this second response was incorrect or no response was given after 20 seconds, the card was shown once more and the participant was asked to copy from it. If it was correct, all correct versions of the word were covered and the participant was again instructed to write the word from memory. The therapist did not give feedback after this third attempt. After each attempt to write the word, the therapist produced the word verbally; however, the participant was instructed not to say the word at any time. After three attempts at writing the word (either copying or writing the word from memory) the therapist proceeded to the next item. The session ended after exactly one hour and the therapist noted which word was the last so the next session could begin with this item.

Multi-Modal Therapy

Figure 4 shows a schematic representation of multi-modal therapy. For each target word the following tasks were completed before the participant progressed to the next word.

1. Semantic distractors task: The participant was shown three written words with similar or associated meanings, one of which was the target word (e.g. painting, picture, art). The therapist said the target word, and the participant was instructed to point to the correct word. The therapist provided feedback on whether the answer was correct.
Regardless of whether it was right or wrong, the participant was then asked to say and then copy the correct word. Feedback was given on the accuracy of the copied word.

2. Phonological task: The participant was asked to listen to the therapist saying three words or non-words and then to pick the word that was different from the other two. A piece of paper consisting of three drawn boxes was placed in front of the participant, each representing a word that the therapist was about to produce. The participant was instructed to point to the box of the word that was different from the other two. The therapist said three words or non-words that sounded similar to each other and pointed to a box for each word. Two of the words, including the middle one were the same. The other word that the participant had to identify as being different was the target word. The therapist gave feedback on whether the choice was correct. The participant was then instructed to say the word and then to write it from memory. The therapist then gave feedback about the accuracy of the written production of the target word. The phonological distractor for this task was a word or non-word with either a substituted phoneme (vowel or consonant) or consonant cluster (e.g. ‘mocolate’ for chocolate or ‘stoctor’ for doctor) or an added or omitted phoneme (e.g. ‘duncle’ for uncle and ‘appoinment’ for appointment). The position of the addition, omission or substitution within the word varied (i.e. word initial, medial or final).

3. Orthographic distractors task: The participant was shown three written words. One of them was the correctly spelt target word. The others were distractors. The therapist said the target word, and the participant was instructed to point to the correct word. The therapist provided feedback on whether the answer was correct. Regardless of whether it was right or wrong, the participant was then asked to say and then copy the correct version of the word. Feedback was not given on the accuracy of the copied word. Distractors were generated by either adding, substituting, omitting or transposing one or two letters. It was, however, still recognisable as similar to the target word.

As in uni-modal therapy, the session ended after exactly one hour. The therapist noted which word was the last to be treated, and the next word was the first to be treated in the next session. At the beginning of the first multi-modal therapy session some practice items were used to ensure that participants understood the tasks.
Post-therapy Assessment

Participants were assessed by the first author on spelling accuracy for all 120 items from multi-modal, uni-modal and control sets directly post therapy (two to four days after the last session) to measure immediate therapy effects, and six weeks post-therapy to establish whether any therapy effects had been maintained. Words from each condition were randomised within the post-therapy list to control for any order effects. If no response was provided within 20 seconds, then the therapist proceeded to the next word.
Results

The results will be set out as follows to directly answer the research questions.

1. Accuracy immediately post-therapy
2. Accuracy of untreated items
3. Accuracy at follow-up

1. Accuracy scores immediately post therapy

Research Question 1: Is a multi-modal therapy more effective than a uni-modal therapy in improving spelling accuracy across matched sets?

Post uni-modal therapy spelling scores

Accuracy scores for all participants are displayed in Figure 5. Four participants (JP, KR, AD & MB) were in Group 1 and received uni-modal as their first treatment. The other participants (DM, JB, SR & EB) were in Group 2 and had uni-modal therapy after multi-modal therapy; therefore the multi-modal words had already been treated (5 weeks previously) at this assessment point. To establish whether uni-modal therapy was effective the scores on uni-modal words were compared to baseline. For all sets, the baseline score was 0/40, as items included into therapy and control sets had to be failed at baseline on two or three occasions. The mean score of the uni-modal sets (22.3) was significantly higher at immediate assessment compared to baseline (Ws+ 0.0, p = .01, 1-tailed). All participants improved significantly on uni-modal sets (McNemar 1-tailed, p < .01 for all participants).

All participants also improved significantly on multi-modal words at the post uni-modal assessment point (Mean: 12.3; Ws+ 0.0, p = .01, 1-tailed), despite the fact that Group 1 had not yet taken part in this therapy, which suggests generalisation to these untreated words for these participants (JP: McNemar 1-tailed, p < .001; KR: McNemar 1-tailed, p = .02 AD: McNemar 1-tailed, p < .001; MB: McNemar 1-tailed, p = .03; DM: McNemar 1-tailed, p < .001; JB: McNemar 1-tailed, p = .002; EB: McNemar 1-tailed, p = .001; SR: McNemar 1-tailed, p < .001).
Post multi-modal therapy spelling scores

Post multi-modal spelling scores for each participant are shown in Figure 6. This was the first therapy for Group 2 (DM, JB, SR, EB) and the second therapy for Group 1 (JP, KR, AD, MB). The mean score of the multi-modal sets (21.3/40) was significantly higher at immediate assessment than at baseline (Ws+ 0.0, p = .01, 1-tailed). All participants improved significantly on multi-modal sets (JP: McNemar 1-tailed, p < .001; KR: McNemar 1-tailed, p < .001; AD: McNemar 1-tailed, p < .001; MB: McNemar 1-tailed, p < .001; DM: McNemar 1-tailed, p < .001; JB: McNemar 1-tailed, p = .01; EB: McNemar 1-tailed, p < .001; SR: McNemar 1-tailed, p < .001).

The mean uni-modal score at this post multi-modal assessment point (13.5) was significantly higher than baseline (Ws+ 0.0, p = .01, 1-tailed), which, again, reflected the performance of all participants (JP: McNemar 1-tailed, p < .001; KR: McNemar 1-tailed, p < .001; AD: McNemar 1-tailed, p = .004; MB: McNemar 1-tailed, p < .001; DM: McNemar 1-tailed, p < .001; JB: McNemar 1-tailed, p = .02; EB: McNemar 1-tailed, p = .002; SR: McNemar 1-tailed, p = .004).
Comparison of multi-modal and uni-modal scores

Figure 7 shows scores on uni-modal words directly after uni-modal therapy compared to scores on multi-modal words directly following multi-modal therapy. The mean scores (22.3/40 for uni-modal and 21.3/40 for multi-modal) were not significantly different from each other (Ws+ 22.0, p = .31, 1-tailed), which was also the case for all participants’ individual scores (JP: $X^2= 0.26$, df= 1, p = .61; DM: $X^2= 0.06$, df= 1, p = .81; KR: $X^2= 2.88$, df= 1, p = .09; AD: $X^2= 0.47$, df= 1, p = .49; JB: $X^2= 0.30$, df= 1, p = .59; SR: $X^2= 0.45$, df= 1, p = .50; MB: $X^2= 0.20$, df= 1, p = .66; EB: $X^2= 4.0$, df= 1, p = .05, 1-tailed).

[Insert Figure 7 about here]

2. Accuracy of untreated items

Research Question 2: Does a multi-modal lead to a greater degree of generalisation to untreated words than a uni-modal therapy?

The mean control score (9.6/40) was significantly higher than baseline immediately post uni-modal (Ws+ 0.0, p = .01, 1-tailed) and multi-modal therapies (Ws+ 0.0, p = .01, 1-tailed), and each participant’s control score improved significantly at both time points (Post uni-modal: JP: McNemar 1-tailed, p < .001; KR: McNemar 1-tailed, p = .01; AD: McNemar 1-tailed, p = .002; MB: McNemar 1-tailed, p = .002; DM: McNemar 1-tailed, p < .001; JB: McNemar 1-tailed, p = .03; EB: McNemar 1-tailed, p = .004; SR: McNemar 1-tailed, p < .001; Post multi-modal: JP: McNemar 1-tailed, p < .001; KR: McNemar 1-tailed, p = .002; AD: McNemar 1-tailed, p < .001; MB: McNemar 1-tailed, p < .001; DM: McNemar 1-tailed, p = .008; JB: McNemar 1-tailed, p = .02; EB: McNemar 1-tailed, p = .002; SR: McNemar 1-tailed, p = .001).

In order to determine whether one therapy resulted in more generalisation than another, mean and individual control scores were compared across therapies (Figure 8). The mean post multi-modal control score (10.8/40) was not significantly higher than the mean post uni-
modal control score (9.6/40) (Ws+ 12.0, p = .22, 1-tailed). For seven participants, individual control scores also did not differ significantly following the two therapies (DM: $X^2 = 1.65$, df= 1, p = .20; KR: $X^2 = 0.08$, df= 1, p = .78; AD: $X^2 = 0.56$, df= 1, p = .46; JB: $X^2 = 0.00$, df= 1, p = .10; SR: $X^2 = 0.00$, df= 1, p = 1.00; MB: $X^2 = 0.07$, df= 1, p = .80; EB: $X^2 = 0.00$, df= 1, p = 1.00, 1-tailed). However, JP had a significantly higher control score following multi-modal therapy than uni-modal therapy ($X^2 = 4.94$, df= 1, p = .03, 1-tailed). As she had multi-modal therapy after uni-modal therapy, it could be that her generalisation following multi-modal therapy was due to the combined effects of uni-modal and multi-modal therapies.

[Insert Figure 8 about here]

It is important to note that after uni-modal therapy the mean uni-modal score was significantly higher than the mean control score (Ws+ 36, p = .01, 1-tailed), indicating an effect of therapy. This reflected the scores of JP ($X^2 = 31.80$, df= 1, p < .001, 1-tailed), KR ($X^2 = 13.31$, df= 1, p < .001, 1-tailed), MB ($X^2 = 7.49$, df= 1, p = .01, 1-tailed), DM ($X^2 = 7.11$, df= 1, p = .01, 1-tailed) and EB ($X^2 = 13.04$, df= 1, p < .001, 1-tailed); however, AD’s, JB’s and SR’s uni-modal scores did not differ significantly from their control scores (AD: $X^2 = 2.76$, df= 1, p = .10; JB: $X^2 = 1.30$, df= 1, p = .26; SR: $X^2 = 0.22$, df= 1, p = .64, 1-tailed). Similarly, after multi-modal therapy the mean multi-modal score was significantly higher than the mean control score (Ws+ 28.0, p = .01, 1-tailed), suggesting a therapy effect. On an individual level this was the case for JP ($X^2 = 8.57$, df= 1, p = .003, 1-tailed), KR ($X^2 = 23.91$, df= 1, p < .001, 1-tailed) and DM ($X^2 = 20.06$, df= 1, p < .001, 1-tailed). The other participants did not perform better on multi-modal words compared to control words (AD: $X^2 = 0.06$, df= 1, p = .81; MB: $X^2 = 2.58$, df= 1, p = .11; SR: $X^2 = 2.58$, df= 1, p = .11; JB: $X^2 = 0.00$, df= 1, p = 1.00; EB: $X^2 = 1.47$, df= 1, p = .23, 1-tailed).

3. Accuracy at follow-up assessment

Research Question 3: Is a multi-modal therapy more effective than a uni-modal therapy in terms of maintenance of learning effects across matched sets?
In order to determine whether any effects of therapies had been maintained, follow-up uni-modal scores were compared to scores on uni-modal words directly following uni-modal therapy and follow-up multi-modal scores were compared to scores on multi-modal words directly following multi-modal therapy. Both the mean uni-modal and the mean multi-modal scores decreased significantly at follow-up (Uni-modal: Ws+ 21.0, \( p = .02, \) 1-tailed; Multi-modal: Ws+ 28.0, \( p = .01, \) 1-tailed), indicating that therapy effects had not been maintained. However, individual results were mixed. DM, EB, and JB maintained both their multi-modal and their uni-modal scores at follow-up (Multi-modal: DM: McNemar 1-tailed, \( p = .13; \) JB: McNemar 1-tailed, \( p = .13; \) EB: McNemar 1-tailed, \( p = .06; \) Uni-modal: DM: McNemar 1-tailed, \( p = 1.00; \) JB and EB: McNemar 1-tailed, \( p = .06). \) Both KR’s uni-modal and multi-modal scores decreased significantly at follow-up assessment (McNemar 1-tailed, \( p < .001 \) for both uni-modal and multi-modal therapy sets). JP and AD’s multi-modal therapy scores were not significantly different at follow-up (JP: McNemar 1-tailed, \( p = .25; \) AD: McNemar 1-tailed, \( p = 1.00): \) however, their uni-modal scores were significantly lower (McNemar 1-tailed, \( p = .02 \) for both). MB’s and SR’s multi-modal scores, on the other hand, did decrease significantly at follow-up (McNemar 1-tailed, \( p = .03 \) for MB and SR), whereas their uni-modal scores did not (MB: McNemar 1-tailed, \( p = .25; \) SR: McNemar 1-tailed, \( p = 1.00). \)

Follow-up control scores were compared to scores on control words both immediately after uni-modal therapy and multi-modal therapy. The mean control score was significantly higher at follow-up (13.5/40) compared to the mean control score immediately after uni-modal therapy (9.6/40; Ws+ 2.5, \( p = .02, \) 1-tailed), but not multi-modal therapy (10.8/40; Ws+ 7.0, \( p = .07, \) 1-tailed). Follow up control scores did not differ for most of the participants when compared to post uni-modal assessment (KR, AD, JB and MB: McNemar 1-tailed, \( p = .50; \) SR and EB: McNemar 1-tailed, \( p = .13) \) or post multi-modal assessment control scores (KR: McNemar 1-tailed, \( p = .50; \) AD: McNemar 1-tailed, \( p = .13; \) MB: McNemar 1-tailed, \( p = .50; \) SR: McNemar 1-tailed, \( p = .06; \) JB and EB: McNemar 1-tailed, \( p = .25), \) indicating that improvements to untreated words were maintained. However, DM’s control score increased to 21/40 from 7/40 after multi-modal therapy and 13/40 after uni-modal therapy, which was statistically significant in both cases (Multi-modal: McNemar 1-tailed, \( p < .001; \) Uni-modal: McNemar 1-tailed, \( p = .004). \) Furthermore, JP’s control scores increased to 29/40 at follow-up (from 14/40 following uni-modal therapy and from 25/40 post multi-modal therapy);
however this score only increased significantly from the post uni-modal control score (McNemar 1-tailed, p < .01).

Figure 9 shows the individual follow-up scores for the three conditions. Similar to immediate post-therapy scores, mean follow-up scores for the uni-modal and multi-modal conditions were not significantly different to each other (Ws+ 22.5, p = .29, 1-tailed). This reflected the results of seven participants (JP: $X^2 = 0.10$, df = 1, p = .76; DM: $X^2 = 0.00$, df = 1, p = 1.00; KR: $X^2 = 0.06$, df = 1, p = .81; AD: $X^2 = 0.06$, df = 1, p = .81; JB: $X^2 = 0.11$, df = 1, p = .74; SR: $X^2 = 0.00$, df = 1, p = 1.00; MB: $X^2 = 1.26$, df = 1, p = .26, 1-tailed). However, EB had a significantly higher score on uni-modal words than multi-modal words at follow-up ($X^2 = 4.17$, df = 1, p = .04, 1-tailed). The mean control score was significantly lower than the mean multi-modal score (Ws+ 20.0, p = .03, 1-tailed); however for individual participants there was no significant difference between these conditions (JP: $X^2 = 1.93$, df = 1, p = .17; DM: $X^2 = 0.45$, df = 1, p = .50; KR: $X^2 = 1.55$, df = 1, p = .21; AD: $X^2 = 0.24$, df = 1, p = .62; JB: $X^2 = 0.14$, df = 1, p = .71; SR: $X^2 = 0.00$, df = 1, p = 1.00; MB: $X^2 = 0.53$, df = 1, p = .47; EB: $X^2 = 0.06$, df = 1, p = .80, 1-tailed). The mean control score was also lower than the mean uni-modal score (Ws+ 28.0, p = .01, 1-tailed), which was true for MB and EB (MB: $X^2 = 4.27$, df = 1, p = .04; EB: $X^2 = 4.17$, df = 1, p = .04, 1-tailed) but not the other six participants (JP: $X^2 = 0.64$, df = 1, p = .43; DM: $X^2 = 0.81$, df = 1, p = .37; KR: $X^2 = 0.59$, df = 1, p = .44; AD: $X^2 = 0.00$, df = 1, p = 1.00; JB: $X^2 = 0.11$, df = 1, p = .74; SR: $X^2 = 0.05$, df = 1, p = .82, 1-tailed).

[Insert Figure 9 about here]
Discussion

A within-participants multiple case study was conducted with eight participants with acquired dysgraphia. Two approaches to spelling therapy were compared: a uni-modal therapy, in which participants copied, covered and recalled written words and a multi-modal therapy, which required participants to select the word from semantic, phonological and orthographic distracters, to say the word, to copy the word and to write the word from memory. The effects of each of these therapies on spelling accuracy were compared to a control (no therapy) condition. All participants had had their stroke at least one year prior to commencement of the study; therefore, it was unlikely that any improvements could be attributed to spontaneous recovery. It was predicted that multi-modal therapy may show a slower learning trajectory than uni-modal, but would be more effective than uni-modal therapy in terms of maintenance of learning and generalisation to untreated control items.

The results showed that all participants improved significantly on treated and untreated words following both therapies compared to baseline. On a group level, effects were not maintained six weeks later. However, on an individual basis, the results were varied; although DM, EB and JB maintained their gains to both therapy sets, there was a significant decrease in spelling accuracy of KR’s uni-modal and multi-modal words, AD and JP’s uni-modal words and MB’s and SR’s multi-modal words compared to immediately post therapy. Control scores were all maintained or increased.

Importantly, all participants expressed a preference for multi-modal therapy due to the variation of tasks. Despite this, there were no significant differences between the effects of uni-modal and multi-modal therapies on spelling accuracy immediately post therapy. Furthermore, a comparison of immediately post-therapy control scores showed no significant differences between the extent to which the two therapies resulted in generalisation to untreated items. JP had larger gains to control items following multi-modal therapy; however, as this was her second therapy these larger gains may be due to the cumulative effects of both therapies. For the majority of participants, neither therapy had an advantage over the other in terms of maintenance of learning. An exception to this was EB. She had a significantly better follow-up score for uni-modal words than multi-modal words. EB had had uni-modal therapy much more recently than multi-modal therapy by the time she completed the follow-up assessment, which might be the reason for her higher scores on uni-modal words. However,
she actually also performed better directly following uni-modal therapy than directly following multi-modal therapy. Although this difference was not quite significant, it is further support that uni-modal therapy was a more successful therapy method for EB. She seemed to benefit from a therapy with more emphasis on writing practice than on improving other linguistic skills.

There are several possible explanations to explain the similarity in effects between the two therapies. Firstly, multi-modality may not be an important enough factor in relearning to outweigh the advantage of more frequent repetitions of a word. The two therapies were matched for session length in order to provide a useful comparison to clinicians about the type of therapy that would provide the most gains when provided within standard therapy sessions. However, as each therapy differed in time spent on each task, this meant that there was a marked contrast between number of items treated per session for each participant and, therefore, the number of times each item was practised throughout the block of therapy. The mean number of words treated per session in uni-modal therapy was 56; whereas the mean number treated in multi-modal therapy was only 32. Therefore, whereas uni-modal words were practised an average of 6.5 times; multi-modal words were only practised an average of 3.6 times across the 5 sessions. It seems likely that the more frequent opportunities to write the word in uni-modal therapy balanced out any expected advantages of using other modalities in multi-modal therapy.

Another reason for the similar results could be that the therapies were not sufficiently distinct from one another. In both therapies, participants saw the word, copied the word, and heard the word. In multi-modal therapy, participants were instructed to say the word, whereas in uni-modal therapy, they were not. However, some participants automatically repeated or read the word as they heard it, although they were discouraged from doing this as it narrowed any differences between the therapies. Moreover, the multi-modal therapy may not have been sufficiently multi-modal as participants completed tasks in different modalities sequentially.

A different interpretation of ‘multi-modal’ could be the simultaneous use of different modalities. For example, people with and without aphasia often communicate ideas by saying and gesturing a word at the same time. Furthermore, in the semantic distractor task, the participants were able to quickly select the spoken word by recognising the correct letters, as they all had sufficient reading ability to do this. For these participants, this might not have led
to any more semantic activation of target words by participants than just hearing the spoken word or looking at the written word, which happened in both therapies. A semantic decision or semantic generation task might have been more successful at strengthening the representation of the target word. Boyle and Coelho (1995) have suggested that semantic tasks requiring participants to generate information about the target may lead to more lasting effects than a more passive semantic task.

Although there were no differences between the effects of the two therapies, there was substantial variation in the performance of individual participants. Some patterns did emerge. Firstly, the participants who made the most gains were those with the lowest pre therapy spelling scores (JP, DM, KR). JP also had a relatively low score on the CAT copying task (Swinburn et al., 2004), indicating impairment in peripheral writing skills, which can be an indicator of a poor response to writing treatment (Beeson, Rising & Volk, 2003). The successful performance of these participants could reflect the fact that they had more room for change. Furthermore, their therapy items were shorter, higher imageability and higher frequency (e.g. guitar, stroke, family, house) which may have been easier to relearn than the therapy items that were selected for the higher level participants (e.g. politician, disagree, Wednesday, interesting) who did not fail these easier items at baseline. Another explanation could be that the participants with more severe language and writing difficulties had a clearer motivation for improving in therapy as they wanted to be able to communicate more effectively (e.g. using writing to support face to face conversations), which led to more effort being put into sessions; whereas less severely impaired participants such as MB or EB could already use their writing skills to send text messages or write a note.

Secondly, some of the higher performing participants showed evidence of the development and use of a strategy. The most noticeable was the strategy used by JP, a participant with surface dysgraphia, who, in previous therapy, had been encouraged to segment target words and to convert phonemes to graphemes. Within the therapies in this study, she segmented words and then remembered the segments the next time she heard them. For example, she remembered ‘chicken’ as ‘chic’ ‘ken’, ‘wife’ as ‘wife’ ‘ee’ and ‘father’ as ‘fat’ ‘her’. In other words, she would store a different phonological representation of the word that helped her to remember the correct orthographic one, so that she would be able to convert these sound segments into written segments. She verbalised these strategies when she first used them, but
was discouraged from speaking about the words during these therapies. However, she reported that she then said these to herself internally after this. She made substantial gains following both therapies and generalisation to untreated words, and personally felt that the increased strategy use and the repetitive use of these strategies on the target words during this time was partly responsible for these gains. DM left spaces within words for letters (usually word-medial) that he could not remember. He then wrote the word again, inserting different letters into the space until he found the correct letter. He used this strategy successfully within therapy and assessment.

This study provided further evidence that lexical therapies can be effective in improving single word writing in people with aphasia, as other studies have shown (e.g. Ball et al., 2001; Beeson, 1999; Clausen & Beeson, 2003; Jackson-Waite et al., 2003; Rapp, 2005). The fact that all participants demonstrated some improvement to untreated control items following both types of therapy was positive. One of the disadvantages to lexical therapies for writing is that they often do not result in generalisation (Beeson & Rapscak, 2002). The exceptions to this have usually been studies in which the participants have a graphemic buffer disorder (Rapp, 2005; Rapp and Kane; 2002; Raymer, Cudworth and Haley, 2003; Sage & Ellis, 2006). In this study, the improvements to untreated items in participants with a range of different dysgraphia types could be attributed to such factors as strengthened phonological, orthographic or semantic systems or a strengthened graphemic buffer (Rapp & Kane, 2002). DM believed that his increase in control item scores at follow-up assessment was due to improved ability to process the spoken word in spelling to dictation tasks (i.e. improved phonological processing). This was a skill that could have improved in both therapies, but was more explicitly encouraged in multi-modal therapy.

Alternatively, participants’ improved control scores could be attributed to general improvements to non-linguistic factors such as effort, attention, motivation or self-monitoring skills. Writing differs to speech in that most people with aphasia, regardless of their severity, will continue to engage in efforts at verbal communication. It is easier to become disengaged from the experience of writing, however, through simple avoidance or delegation of writing tasks. The majority of the participants in this study had not attempted to write very often since their stroke, which may explain why during a period of increased writing and increased effort, they demonstrated some generalised improvements. The participants who made the
most substantial improvements to untreated items were JP and DM. In both cases, it is likely that they improved their strategy use throughout therapy and were able to use these on untreated words. JP, DM and KR all reported that they noticed improvements when trying to complete everyday writing tasks, such as emailing or writing shopping lists and that they had been writing more often since therapy started. This could further explain their improvement to control items. Again, these participants had been learning functional, high frequency words, such as names of family members, which were likely to be useful in everyday writing activities.

A limitation of this study was that both therapies and assessments were administered by the first author; therefore blinding was not possible. According to Tate, McDonald, Perdices Togher, Schultz & Savage (2008), using the same person to provide assessment and therapy introduces a risk of observer bias into a study. A further limitation of this study was that with relatively small numbers it has not been possible to conduct correlation analyses to investigate whether therapy success can be predicted by the nature of a participant’s spelling, language or cognitive impairment, as has been the case in the anomia literature, where studies have shown that participant performance in therapy can be predicted from cognitive and/ or linguistic profiles (e.g. Lambon Ralph, Snell, Fillingham, Conroy & Sage, 2010). Future studies should use larger numbers so that individual factors can be investigated. Clinicians will then be able to use this information to determine which patients will benefit from certain therapies.

Overall, the findings have clinical implications in that they have suggested that relatively brief episodes of simple behavioural treatments and practice can be effective in improving spelling accuracy in adults with a range of linguistic and spelling impairments. An interesting observation was that where there was flexibility within therapy tasks, participants may often initiate strategies and make proactive use of their processing strengths to find ways of enhancing and maintaining spelling accuracy.
References


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Figure 1. The Triangle Model (after Plaut, McClelland, Seidenberg & Patterson, 1996)

Figure 2. Study design
Figure 3. Uni-modal therapy
Multi-modal Therapy

Semantic distracter task, say word, copy word

Phonological distracter task, say word, write word

Orthographic distracter task, say word, copy word

Next word

Figure 4. Multi-modal therapy
Figure 5. Post uni-modal therapy spelling accuracy scores

Figure 6. Post multi-modal therapy spelling accuracy scores
Figure 7. Directly post therapy spelling accuracy scores for uni-modal and multi-modal word lists

Figure 8. Spelling accuracy scores of untreated control items
Figure 9. Spelling accuracy scores at 6 week follow-up

Number of words correct

Participant

JP DM KR AD JB SR MB EB Mean

Uni-modal words
Multi-modal words
Control

*p < .05
Table 1. Demographic Data and Screen Scores

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Table 2. BDAE and PPT Scores

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Table 3. PALPA Scores

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