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Computer-Based Naming Treatment for Semantic Variant Primary Progressive Aphasia With History of Traumatic Brain Injury: A Single-Case Experimental Design

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ABSTRACT

Purpose: The purpose of this study was to investigate the effectiveness of a self-administered naming treatment for one individual, B.N., presenting with semantic variant primary progressive aphasia (svPPA) and a history of traumatic brain injury (TBI).

Method: Naming treatment included components of Lexical Retrieval Cascade Treatment and was self-administered using an adaptive spaced retrieval software, Anki. Using a multiple-baseline, single-case experimental design, naming accuracy probes were taken during pretreatment, treatment, posttreatment, and follow-up (through 12 months) for 60 trained words and 10 untrained words. Item-level Bayesian generalized mixed-effects models were used to estimate (a) the treatment effect for trained words, (b) change in untrained words, and (c) maintenance of treatment effects from posttreatment to each subsequent follow-up.

Results: Statistical analyses revealed that a gain of 35 out of 60 trained words (35.3; 90% CI [30.6, 39.5]) was directly attributable to treatment. Following treatment, evidence of generalization to untrained words was not observed. During the follow-up period, there was gradual decline in naming accuracy of trained items.

Conclusions: The positive treatment results reported here support the use of self-administered naming treatments for those with svPPA and a history of TBI. Although the utility of this treatment approach is constrained by patient factors including motivation, self-administered naming treatments represent a unique opportunity to expand access to speech-language intervention for people with svPPA, including those with concomitant diagnoses.

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Primary progressive aphasia (PPA) is a neurodegenerative disorder that predominantly affects speech and/or language functions (Mesulam, 2001). Individuals living with PPA initially present with deficits in their ability to communicate. As the disorder progresses, impairments in speech and language functions become gradually more pronounced and individuals may also show declining cognitive and motor functioning (Rogalski & Mesulam, 2009).

International consensus criteria identify three PPA variants: nonfluent/agrammatic variant (nfvPPA), logopenic variant (lvPPA), and semantic variant (svPPA), each with a unique array of behavioral features, pattern of underlying brain atrophy, and neuropathological basis (Gorno-Tempini et al., 2011; Montembeault et al., 2018). In the current study, we investigated the effects of treatment for an individual with svPPA and therefore will focus the following literature review primarily on svPPA,

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though many of the treatment studies referenced include participants across PPA subtypes.

Also referred to as semantic dementia (Mesulam et al., 2009), svPPA is characterized by impaired semantic memory leading to anomia and word comprehension deficits (Gorno-Tempini et al., 2011; Hodges & Patterson, 2007; Hurley et al., 2012). Individuals with svPPA also demonstrate loss of object knowledge (Gorno-Tempini et al., 2011; Hodges & Patterson, 2007). Repetition and speech motor abilities, on the other hand, remain relatively intact (Gorno-Tempini et al., 2011; Montembeault et al., 2023). The loss of conceptual knowledge characteristic of svPPA correlates with left-predominant atrophy of the anterior temporal lobe (Gorno-Tempini et al., 2011; Hodges & Patterson, 2007; Hurley et al., 2012; Spinelli et al., 2017) and is associated with TDP-43 proteinopathy at autopsy (Spinelli et al., 2017). Due to a decline in conceptual knowledge, people with svPPA experience a gradual loss of independence (Hardy et al., 2023). Person-centered interventions for svPPA aim to increase individuals' ability to participate meaningfully in daily life and may include the training of personally meaningful words (Rogalski & Khayum, 2018).

The body of evidence supporting the value of speech-language interventions for individuals affected by PPA, including svPPA, has grown substantially over the last 2 decades (Cadório et al., 2017; Carthery-Goulart et al., 2013; Cotelli et al., 2020; Volkmer, Rogalski, et al., 2020; Wauters et al., 2023). Even so, the PPA treatment literature remains relatively modest compared to that for stroke-induced aphasia. Additional research is needed to investigate important clinical questions, such as the relative benefits of different treatment delivery methods, the appropriate use of technology in supporting communication, optimal dosage over the course of disease progression, and the impact of concomitant neurological disorders on response to treatment.

Naming Treatments for PPA

Speech-language intervention studies for PPA have most often focused on the relearning of vocabulary through repeated practice and typically involve training the association between pictures and corresponding word forms (written and/or verbal). These interventions have proven to result in successful relearning of trained words (for reviews of PPA naming treatments, see Cadório et al., 2017; Carthery-Goulart et al., 2013; Cotelli et al., 2020; Volkmer, Rogalski, et al., 2020; Wauters et al., 2023) with at least some long-term maintenance possible, particularly with ongoing practice of targeted words (Cadório et al., 2017). Among the PPA variants, individuals with svPPA may be less likely to show generalization of learning beyond trained items (Cadório et al., 2017; Jokel, 2019). It has been proposed that, because svPPA directly affects semantic memory, individuals with svPPA rely more heavily on episodic memory to support new learning, limiting their ability to generalize beyond a specific learning context (Bier et al., 2009). However, generalization has recently been documented in individuals in the mild-to-moderate stages of svPPA following the administration of treatments that include training of self-cueing strategies (Beales et al., 2016, 2021; Grasso et al., 2021; Henry et al., 2013, 2019).

In strategy training approaches, the aim of treatment is to promote self-cueing to improve word retrieval (Beales et al., 2016, 2021; Grasso et al., 2021; Henry et al., 2013, 2019; Newhart et al., 2009; Savage et al., 2013, 2015a). During treatment, patients actively recall residual semantic and/or word-form knowledge for target words. One such treatment, initially developed at the University of Arizona, is Lexical Retrieval Cascade Treatment (LRT; Dial et al., 2019; Grasso et al., 2021; Henry et al., 2013, 2019). LRT includes a series of tasks to prompt retrieval of information about target words, including semantic feature analysis (Boyle & Coelho, 1995) as well as cues to retrieve residual phonemic and orthographic information. Because this approach employs embedded strategy training, it may have a greater chance of eliciting generalization to untrained items. In fact, for svPPA participants, clinician-administered LRT has resulted in significant gains in naming accuracy for trained items through 1 year posttreatment (Grasso et al., 2021; Henry et al., 2019) as well as generalization to untrained items through 6 months posttreatment (Henry et al., 2019).

Self-Administered Treatment for svPPA

Despite a growing evidence base for speech-language interventions, people with PPA often encounter limited access to services due to underreferral, a lack of available clinicians familiar with PPA, and limited reimbursement from insurance (Riedl et al., 2014; Taylor et al., 2009; Volkmer, Spector, et al., 2020). Self-administered, computer-based treatment programs can provide a treatment option for people who have limited access to traditional face-to-face services. Because cognitive abilities are relatively spared in the initial stages of PPA, individuals are often able to independently operate computers and other technology, making self-administered treatment a viable option for this population (Jokel et al., 2014).

Although a number of studies include some computer-based practice, only a handful of studies have investigated the effects of self-administered computer-based naming treatment options for svPPA (Evans et al., 2016; Heredia et al., 2009; Krajenbrink et al., 2020; Lavoie et al., 2020; Savage et al., 2014, 2015a). As is typical of noncomputerized naming interventions for svPPA, these studies have utilized an approach that involves repeated practice of target words (written and/or spoken) in conjunction with pictured stimuli. Evans et al. (2016) added the presentation of semantic information along with each target word, and Lavoie et al. (2020) included a task in which participants responded to yes/no questions querying semantic features of target words. Each of these self-directed computer-based interventions resulted in a significant increase in naming accuracy for trained words. Of the studies that reported on maintenance, most described some degree of maintained gains in follow-up probes (Heredia et al., 2009; Lavoie et al., 2020; Savage et al., 2015a) with one study reporting a return to baseline after treatment ended (Krajenbrink et al., 2020). Lavoie et al. also observed generalization of gains to untrained words for one of two participants with svPPA. Otherwise, generalization to untrained stimuli was either not reported or not observed (Evans et al., 2016; Heredia et al., 2009; Krajenbrink et al., 2020; Savage et al., 2014, 2015a).

Spaced Retrieval Treatment and Its Utility in svPPA

Spaced retrieval training (Haslam et al., 2011; Landauer & Bjork, 1978) is an additional treatment approach that has been used to promote relearning of words for people with svPPA (Bier et al., 2009; Evans et al., 2016; Meyer et al., 2018; Reilly, 2016). During a traditional spaced retrieval training session, the learner is prompted to recall information, at either equally spaced intervals or incrementally longer intervals with each accurate response. After each response, they receive feedback regarding accuracy. Incremental or distributed testing is intended to result in increased retention over time.

Spaced retrieval training has proven effective for individuals with memory or language impairments of various etiologies (Karpicke, 2017), including dementia (Abrahams & Camp, 1993; Balota et al., 2006; Brush & Camp, 1998; Haslam et al., 2011), traumatic brain injury (TBI; Haslam et al., 2011; Sumowski et al., 2010), and stroke-induced aphasia (Brush & Camp, 1998; Fridriksson et al., 2005; Friedman et al., 2017; Middleton et al., 2016; Quique et al., 2022). In one study that compared learning strategies in healthy adults, patients with acquired brain injury, and patients with dementia, researchers found that participants had greater retention of information using errorless learning and spaced retrieval compared to trial-and-error learning, with the greatest retention for information learned through spaced retrieval treatment (Haslam et al., 2011).

There are differing accounts as to why spaced retrieval is an effective means of learning or relearning words (Karpicke, 2017). One theory posits that when a learner is prompted to recall a particular word, they activate a network of semantically related words (Carpenter, 2009). By strengthening this network of associations, the learner increases the likelihood of recalling the word on future attempts. This "elaborative retrieval" hypothesis aligns well with theories of learning that underlie traditional speech-language treatments such as Semantic Feature Analysis, in which a patient is asked to describe characteristics of a target word (Boyle & Coelho, 1995) with the aim of strengthening semantic association networks for the target. Others have suggested that minimizing errors, thereby minimizing errored learning, is a key component of successful spaced retrieval treatments (Bier et al., 2009; Fridriksson et al., 2005).

Several studies have included a spaced retrieval component in treatment for individuals with svPPA (Bier et al., 2009; Evans et al., 2016; Meyer et al., 2018; Reilly, 2016) with the aim of supporting learning in the context of impaired semantic memory. Bier et al. (2009) directly investigated the efficacy of spaced retrieval, comparing two treatment approaches in a single participant with svPPA: (a) simple repetition combined with formal-semantic therapy and (b) spaced retrieval combined with formal-semantic therapy. Both approaches contributed to significant gains in naming trained words. While the participant demonstrated greater accuracy and fewer semantic errors in the spaced retrieval condition, these differences did not reach statistical significance.

In a more recent study, Evans et al. (2016) provided naming therapy to an individual with svPPA using Anki, a free software program. Rather than the more rigid distribution schedule used in traditional spaced retrieval training, Anki utilizes an adaptive algorithm to optimize learning by presenting difficult, low-accuracy words more frequently than high-accuracy words (Woźniak & Gorzelanczyk, 1994). Treatment was home based and self-directed, and the participant reported that she enjoyed working on word finding at home. After the 20-month treatment period, during which the participant showed a decline in her overall ability to retain semantic information, she successfully learned 139 of 591 trained words. The majority of these words were learned during the 1st year when the participant's semantic memory was less impaired. These results demonstrate the utility of an adaptive spaced retrieval approach during the early stages of disease progression as a viable means for individuals with svPPA to relearn lost words with little clinician involvement.

Concomitant svPPA and TBI

The literature to date investigating the effects of treatment in PPA has comprised largely homogenous samples that exclude individuals presenting with concomitant neurological disorders. Therefore, while clinicians will inevitably encounter clients with complicated neurological histories, little is known about how concomitant neurological conditions may impact the effectiveness of treatments that have previously proven effective in PPA.

Exploring treatment for people with svPPA and a history of TBI may be of particular importance. Though the causal relationship is unclear, svPPA and postconcussion syndrome co-occur at a relatively high rate. According to data from the National Alzheimer's Coordinating Center, a history of severe TBI (defined as the loss of consciousness for over 5 min) increases the risk of frontotemporal dementia (FTD) by approximately 67% (Deutsch et al., 2015), suggesting that brain injury may act as an accelerant or catalyst for the later development of FTD syndromes (LoBue et al., 2016). Clinicians working with individuals who have PPA may therefore face an increased rate of previous severe TBI compared to other clinical populations. As such, there is a need to characterize the effects of speech-language interventions administered to individuals with PPA and a positive history of severe TBI.

Current Study

The overarching purpose of this study was to explore the utility of a self-administered naming intervention for one individual, B.N. (fictitious initials), who presented with both svPPA and a history of severe TBI. The self-administered naming treatment included self-cueing strategic training, which has proven effective in treating anomia for individuals with svPPA (Grasso et al., 2021; Henry et al., 2013, 2019), and spaced retrieval, a learning method that has previously been used to improve learning and retention of information in both svPPA (e.g., Evans et al., 2016) and TBI (e.g., Sumowski et al., 2010). In the current study, we assessed the participant's naming accuracy for both trained and untrained words before treatment, immediately after treatment, and during three follow-up sessions at 3, 6, and 12 months posttreatment. Our research questions and hypotheses were as follows:

Research Question 1. Will the participant improve naming accuracy for trained words after completing a self-directed spaced retrieval treatment with strategic practice?

We hypothesized that, despite the possible long-standing effects of B.N.'s previous TBI, the participant would respond positively to naming treatment, demonstrated by a meaningful increase in naming accuracy on the 60 trained words from pre- to posttreatment.

Research Question 2. Will strategic retrieval of semantic and word-form information lead to generalization of treatment gains to untrained words?

Studies including strategy training with clinician-led treatment have reported increased accuracy for untrained items in some participants with svPPA (Beales et al., 2016, 2021; Henry et al., 2019). On the other hand, most studies of self-administered computer-based treatments for svPPA (Evans et al., 2016; Heredia et al., 2009; Krajenbrink et al., 2020; Savage et al., 2014, 2015a) have not reported a generalization effect for untrained stimuli. We explored the possibility that B.N. would demonstrate generalization to untrained words by evaluating change in performance on 10 untrained words from pre- to posttreatment.

Research Question 3. Will the participant maintain treatment gains during the follow-up period with continued practice?

In previous studies utilizing strategic self-cueing training for individuals with svPPA, maintenance of treatment effects for trained words has been observed up to 12 months posttreatment (Grasso et al., 2021; Henry et al., 2019). Therefore, we hypothesized that, with continued practice, B.N. would maintain treatment gains for up to a year posttreatment, demonstrated by relative stability in naming accuracy on trained words from posttreatment to follow-up time points (at 3, 6, and 12 months posttreatment).

Method

Participant

The participant in the current study, B.N., was a right-handed, monolingual English-speaking man who was 62 years of age at the time of enrollment. He was a business owner and manager with a bachelor's degree. B.N. received speech-language treatment in 2017 through the Aphasia Research and Treatment Lab at The University of Texas at Austin. Study procedures were approved by the institutional review board at The University of Texas at Austin, and the participant gave written informed consent for participation.

At the time of the study, the participant had a diagnosis of svPPA in addition to a complex neurological history including a severe TBI. In 1985, B.N. fell approximately 30 ft from the roof of his home and sustained a closed-head TBI, resulting in a right temporal lobe contusion. He was unconscious for an unknown period of time and spent 10 days in the hospital. During recovery, B.N. exhibited prominent cognitive and behavior changes, including increased agitation. He is reported to have recovered from the majority of these initial symptoms and eventually stabilized, although his spouse reported that he continued to be more socially reserved and spoke more slowly relative to his premorbid status. B.N. was able to return to his typical activities of daily living, including managing a successful business.

In 2014, B.N. experienced hearing changes in his left ear, for which he later began using a hearing aid. While investigating this change in hearing, his medical team also discovered a benign pituitary mass, which they surgically removed. A magnetic resonance imaging (MRI) at that time showed a loss of brain tissue in the right temporal region, the orbitofrontal cortex, and the posterior parietal cortex, attributed to the late effects of TBI (see Figure 1).

Between 2014 and 2016, B.N. began to experience progressive language decline, specifically in the domains of word finding and word comprehension. A clinical MRI scan obtained in 2016 (see Figure 1) revealed additional atrophy of the left anterior temporal lobe compared to the 2014 scan. A positron emission tomography scan at that same time revealed bilateral hypometabolism in the anterior temporal lobes, which was more prominent in the left hemisphere, as well as hypometabolism in the right parietal lobe. B.N. received a diagnosis of svPPA after language testing revealed impairments in naming and single-word comprehension with preserved repetition, motor speech, and grammar.

For the current study, a brief cognitive–linguistic battery was administered prior to the introduction of treatment. B.N.'s performance on this battery (presented in Table 1) revealed marked anomia and word comprehension difficulties. B.N. presented with impaired confrontation naming on the Boston Naming Test (Kaplan et al., 2001) with a score of 10/60 and the Western Aphasia Battery–Revised (WAB-R; Kertesz, 2006) with a score of 7.6/10. B.N. correctly identified eight of 16 words on a subset of items from the Peabody Picture Vocabulary Test (Dunn & Dunn, 1997; Kramer et al., 2003), indicating deficits in single-word comprehension; however, performance on a shortened version of the Pyramids and Palm Trees Test (Howard & Patterson, 1992) was relatively spared. WAB-R scores for fluency (9/10), information content (9/10), comprehension (10/10), and repetition (10/10) indicated relatively intact abilities, and B.N.'s ability to repeat digits, an indicator of phonological working memory (Gorno-Tempini et al., 2008), was within normal limits for backward repetition and just below normal limits for forward repetition. Connected speech was fluent, grammatically intact, and without motor speech impairment. B.N. also performed relatively well on a cognitive screening tool, as indicated by a score of 27/30 on the Mini-Mental State Exam (MMSE; Folstein et al., 1975).

Taken together, pretreatment assessment results indicated deficits in naming and single-word comprehension in the context of relative sparing of other speech-language and cognitive abilities. This pattern of performance was aligned with current consensus criteria for a diagnosis of svPPA (Gorno-Tempini et al., 2011).

Experimental Design

During initial meetings with the study clinician, B.N. expressed interest in a self-administered and home-based approach, rather than a clinician-administered intervention. In response, we designed a self-directed treatment in which B.N. used the free flashcard software Anki to target naming of personally relevant nouns. We used a single-case, multiple-baseline design to evaluate the effects of intervention. The goals of this intervention were to (a) improve naming for a select set of personally relevant

Figure 1. Magnetic resonance imaging results from 2014 and 2016 showing damage to the right anterior temporal region secondary to traumatic brain injury and increased atrophy in the left anterior temporal region in T2 axial plane. L = left; R = right.

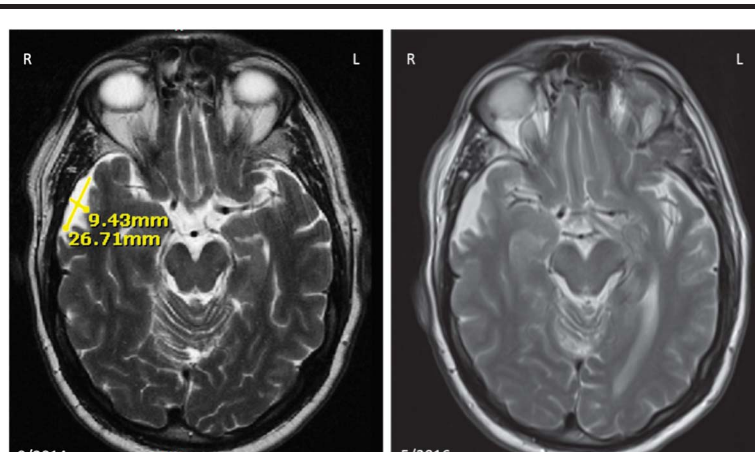


Table 1. Performance on a cognitive–linguistic battery administered to B.N. at each time point.

Assessment	Pre	Post	3-month follow-up	6-month follow-up	1-year follow-up	Normative data *M* (*SD*)
Boston Naming Test (/60 = long version; /30 = alternating short versions A and B)	10/60	10/60	4/30 (A)	4/30 (B)	2/30 (A)	55.6/60 (3.5)[a] 26.7/30 (2.8)[b]
Western Aphasia Battery						
Information Content (10)	9	9	9	9	10	
Fluency (10)	9	9	9	9	9	
Comprehension (10)	10	10	10	9.9	9.8	
Repetition (10)	10	9.8	9.9	10.0	9.4	
Naming (10)	7.6	8.3	7.0	6.3	6.3	
Aphasia Quotient (100)	91.2	92.4	89.6	88.4	89.2	> 93.8
Peabody Picture Vocabulary Test (16)	8	9	10	10	9	
Pyramids and Palm Trees Test (14)	13	14	14	14	11	13.9 (4.5)[c]
MMSE (30)	27	26	24	24	23	29 (1.3)[d]
Digit Span Forward	6	6	5	5	4	7.2 (.9)[e]
Digit Span Backward	6	4	4	5	5	5.4 (1.3)[e]

Note. Boston Naming Test (Kaplan et al., 2001); Western Aphasia Battery–Revised (Kertesz, 2006); Peabody Picture Vocabulary Test (Dunn & Dunn, 1997; Kramer et al., 2003); Pyramids and Palm Trees Test (Howard & Patterson, 1992); Mini Mental State Exam (MMSE; Folstein et al., 1975).

[a]Normative data for ages 60–64 years from Tombaugh and Hubiey (1997). [b]Normative data for ages 66–75 years, 13–16 years of education, from Saxton et al. (2000). [c]Normative data from Breining et al. (2015). [d]Normative data for ages 60–64 with college experience from Crum et al. (1993). [e]Normative data from University of California, San Francisco Memory and Aging Center Bedside Neuropsychological Screen (Henry et al., 2013; Kramer et al., 2003).

words using spaced retrieval and (b) increase the participant's use of word-finding strategies.

Prior to treatment, clinicians provided training and written instructions during an in-person session to familiarize the participant with the computer program. The clinicians also ensured that Anki was installed and functional on the participant's computer and that B.N. was able to operate the program independently. Following this initial session of training, B.N. was responsible for conducting his own treatment sessions for 30 min daily over a 7-week treatment period. Because the treatment was based on principles and treatment steps of LRT, we prescribed a similar dosage to that reported in previous studies utilizing that treatment (e.g., Henry et al., 2019). The 30-min duration of sessions was also in line with the only documented case of a participant with svPPA using Anki to practice naming (Evans et al., 2016). B.N. was also instructed on how to use the "Study Ahead" feature in Anki, which allows for continued practice after all items are correctly named, to ensure that he was able to practice for the entire 30-min period. Adherence to the prescribed schedule was tracked by the Anki program; however, other aspects of the treatment, such as written practice of targets (see below), were only checked briefly during weekly probe sessions. The participant was asked to hold up a paper where he had written words during practice as a means to ensure that he was consistent in completing written practice. The participant was observed to be consistent with completing written practice throughout therapy.

The target nouns for this study (60 trained and 10 untrained) were familiar and meaningful to the participant. Images that were provided by the participant and his spouse were prioritized for inclusion across trained and untrained sets and were supplemented by personally relevant stock photos to ensure that an adequate number of items were eligible for inclusion. For each of the 70 words, B.N. labeled the word incorrectly on at least two of three baseline probes, establishing a baseline accuracy at or near zero for each set of words. Nouns that met inclusion criteria were assigned across seven sets of 10 words (six trained sets and one untrained set) with closely related words (either phonologically or semantically similar) placed in separate groups. Each set of words was balanced for overall frequency, word length, familiarity, and imageability using data from the Corpus of Contemporary American English (Davies, 2009) and the Medical Research Council Psycholinguistic Database (Coltheart, 1981; Wilson, 1988).

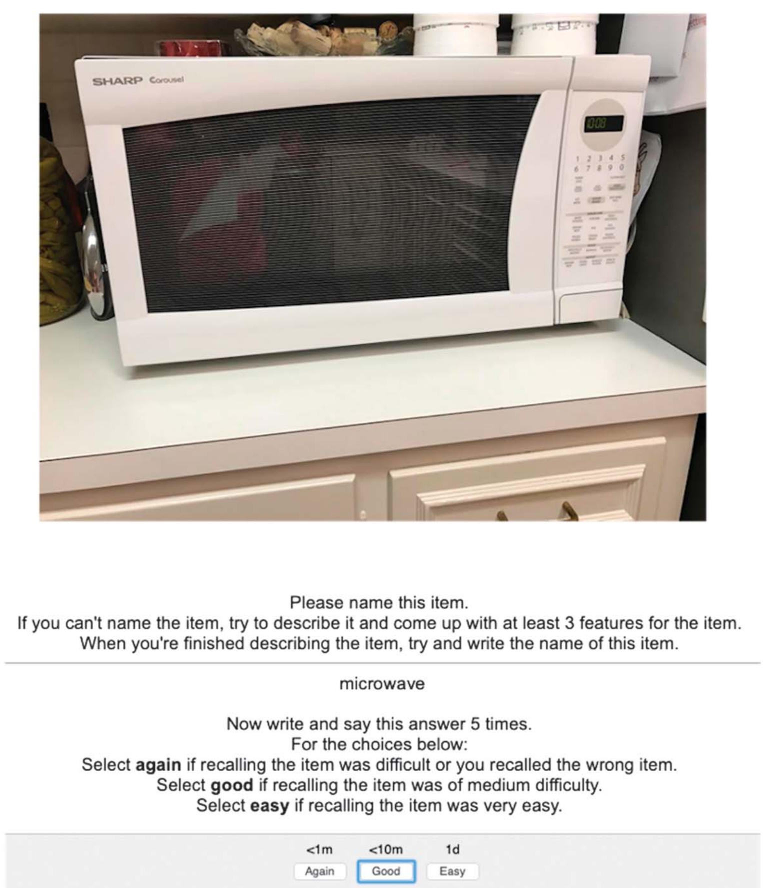
As previously mentioned, the 7-week intervention was largely based upon LRT (Henry et al., 2013, 2019), a treatment approach designed to improve naming of target items through the recall of residual semantic, episodic, orthographic, and phonological information and to promote the use of self-cueing strategies during instances of word-finding difficulty. Due to the nature of the self-administered treatment in the current study, we abbreviated the steps and modified the prompts provided by the program. One such modification was to specify that the participant should only recall residual information about (i.e., describe features of) items that could not be named. In the absence of clinician coaching of strategy use, this focus on unnamed words was meant to implicitly encourage intentional use of self-cueing in moments of word-finding difficulty. During the 30-min practice sessions, B.N. rehearsed each word in the current training set using Anki's digital flashcards and read the following prompts on each flashcard:

1. Please name this item.
2. If you can't name the item, try to describe it and come up with at least three features for the item.
3. When you're finished describing the item, try and write the name of this item.
4. Click the picture to reveal the word.
5. Write and say the answer 5 times.

After completing each step for an individual item (see example in Figure 2), B.N. had the option of choosing one of three buttons at the bottom of the Anki desktop window, depending on self-perceived ability to recall the word. If he perceived that it was very difficult to recall the label or if he was unable to accurately remember the word, he would click "again." If he experienced some difficulty recalling the word but did so accurately, he would click "good." If the item was very easy to recall, he would click "easy." These judgments about the accuracy and ease of recall were considered by the Anki algorithm, which varied the interval between presentations according to difficulty. Only words that were "easy" to recall were taken out of the rotation until the next practice session.

Each week, the clinician met with B.N. using the video-conference software Fuze to probe performance (i.e., naming accuracy) on each of the 60 trained/to-be-trained words and 10 untrained words. Following performance probes, the set of words that was trained during the previous week was then added to a "Mastery Deck" and a new set of 10 words was introduced into treatment. This staggered introduction of word sets into treatment provided the multiple-baseline design of the study, with potential for replication of treatment effect across trained sets.

Figure 2. Screenshot of a practice item on Anki with prompts to retrieve residual information.



B.N. was instructed to practice the items in the Mastery Deck for 15 min after completing the set-in-training. The Mastery Deck was used to control which items the participant practiced each week; namely, he would first practice the items in training before practicing previously introduced items. However, clinical use of Anki for naming treatment would not require a Mastery Deck as Anki automatically adds new words into the main deck so that all items can be practiced according to level of difficulty.

Following the treatment period and posttreatment probes, B.N. continued to practice words from the Mastery Deck, which then included all 60 trained words. In order to simulate home practice that may be provided by a speech-language pathologist in the field, he was encouraged to practice the deck regularly to maintain naming accuracy. We tracked the number of sessions practiced during and after the formal treatment period using data generated by the Anki program.

The primary outcome measure for the study was naming accuracy on sets of trained and untrained words. Following three pretreatment probes, probes were collected each week (total of 7 weeks) during treatment. The treatment period was followed by two posttreatment probes, which occurred on the 3rd and 4th days after treatment ended. Additionally, a single set of probes was collected at each follow-up (3, 6, and 12 months posttreatment).

Self-Assessment of Change

We administered an in-house posttreatment survey (Henry et al., 2013, 2019) to the participant to assess functional changes in his communication. The survey included questions about his performance in treatment as well as the impact of treatment on everyday communication. To respond to each question, B.N. chose a number on a 7-point Likert scale ranging from *a lot worse* (−3) to *a lot better* (3). We used this scale to assess B.N.'s perception of how his communication had changed in several areas including naming, spelling, fluency, and self-monitoring skills as well as the level of frustration, comfort, and confidence he experienced with his spouse and other communication partners.

Statistical Method

Overview of analyses. We used item-level Bayesian generalized mixed-effects models to evaluate the effects of the self-administered naming intervention on naming performance. A single model estimated the effects of introducing words into treatment and an overall treatment effect size (Research Question 1) for the six sets of trained words. This same model was also used to investigate possible generalization (Research Question 2) in the untrained Set 7. A separate model was then used to estimate maintenance of treatment effects (Research Question 3) from the end of the treatment to three follow-up time points (at 3, 6, and 12 months posttreatment). Both models used item-level accuracy (correct or incorrect) as the dependent variable, with a Bernoulli probability distribution and a logit link function. Analyses were conducted in R (R Core Team, 2019) using cmdstan (Stan Development Team, 2021) accessed via the brms package (Bürkner, 2017). Statistical methods for each research question are outlined below. In describing the data set and analysis, we refer to all time points before the participant began practicing a given set of words as the *baseline phase* and all time points after the introduction of a set of words to treatment as the *treatment phase*. Additional detailed descriptions including prior distributions, model fitting, and model evaluation are reported in Appendixes A and B. R code is reported in Supplemental Material S1 and a sensitivity analysis is reported in Supplemental Material S2.

Estimating treatment effect. To evaluate treatment effects, we used an interrupted time series approach as described by Huitema and Mckean (2000), Moeyaert et al. (2017), and Cavanaugh et al. (2023). This approach is well suited for analyzing single-case, multiple-baseline design data, estimating (a) performance before the onset of the baseline phase (i.e., the intercept), (b) any trends in performance before the introduction of an intervention (i.e., the baseline slope), (c) the change in naming accuracy immediately after the intervention begins (i.e., the level change), and (d) the difference in trend between the baseline phase and the treatment phase (i.e., the slope change). This model included data from the six trained sets (Sets 1–6) and the untrained set (Set 7). While data from Sets 1–6 were used to estimate the baseline slope, level change, and slope change, data from Set 7 were only included in the estimate of the baseline slope and did not influence the estimate of level change or slope change. Because group-level effects permitted each set to have its own intercept, baseline slope, level change, and slope change, this approach allowed us to include data from Set 7 to better estimate the baseline slope while obtaining treatment effects specific to Sets 1–6.

To estimate an overall effect size for trained words, we took the difference between the model's posterior distributions for the final treatment baseline probe of each trained word (Sets 1–6), resulting in an estimated number of words gained with an associated 90% credible interval (CI). This approach reflects the structure of the data in that words are nested into sets, with a different number of baseline and treatment phase probes designated to each word and each set of words (i.e., words within sets introduced in a staggered fashion). For example, B.N. did not begin practicing Set 2 until after the fifth performance probe. To calculate the effect size for the words trained in Set 2, we subtracted B.N.'s estimated performance for these words at the fifth probe from his estimated performance at the final treatment probe.

Investigating generalization to untrained words. Because the interrupted time series model included group-level intercepts for words nested within sets and corresponding group-level slopes for each of the three main population-level effects (baseline slope, level change, and slope change), we were able to estimate a baseline slope specific to Set 7. Using this set-specific baseline slope, we estimated an overall effect size for Set 7 by taking the difference in posterior distributions between the end of treatment and the last probe session preceding any treatment (Session 4) for Set 7. This approach yielded a comparable effect size in the number of words gained with an associated 90% CI.

Assessing maintenance of gains relative to posttreatment. A separate model structure was used to determine how well B.N. maintained naming accuracy during the follow-up period for Sets 1–6. This model included a four-level categorical population-level effect of time point (posttreatment and 3-, 6-, and 12-month follow-ups) with posttreatment set as the reference level. Therefore, model parameters describe the change in performance from the end of treatment to each follow-up time point. Group-level effects included intercepts and slopes for words nested within sets.

Results

Adherence to Treatment

B.N. attended all scheduled probing sessions virtually and completed naming accuracy probes, including all posttreatment and follow-up probes. He engaged in home practice on 90% of days during the 7-week treatment period. Data reported by the Anki program indicated that B.N. did not consistently adhere to the recommended 30-min practice time, averaging 10.9 min per session (range: 1.3–30.1) with the program for 8 total hours of practice during the intervention period. From the end of treatment through follow-up probes, B.N.'s practice decreased notably. He practiced 6–7 times between each follow-up probe and logged a total of 1 hr and 43 min of training with the Mastery Deck of 60 words during the entire 12-month follow-up period.

Multiple-Baseline Data

Visually, the data (see Figure 3) show a clear overall effect of treatment, evidenced by the relatively flat performance across sets before they are introduced to treatment, an improvement in performance after each set is introduced to treatment, and then continued high performance through posttreatment probes. Small gains can be observed across sets during the baseline measurements. However, only Set 5 showed a clear upward trend before the set was introduced to treatment. Notably, at posttreatment, B.N. achieved accuracy on only one of 10 words in Set 7, the set of untrained words.

Estimating Treatment Effect

Using the Bayesian interrupted time series model (see results of the model in Table 2 below), we calculated the difference between performance at posttreatment and performance on each set of trained words directly prior to its introduction to treatment. The resulting overall effect size estimate was 35 words (35.3; 90% CI [30.6, 39.5]) out of 60 trained words. Analysis of the interrupted time series model (see Table 2) indicated that the estimated effect of intercept across all sets was −3.70 (90% CI [−4.85, −2.72]), meaning the probability of a correct response at the beginning of the study was approximately 2%. A starting accuracy of 2% is consistent with observed performance as words were selected for treatment only if they were incorrectly named in at least two of three pretreatment trials.

The estimated effect of baseline slope was 0.23 (90% CI [0.00, 0.47]), indicating that the odds of a correct response increased by a magnitude of 1.26 at each probe during the baseline phase. This estimate represents a modest rise in naming accuracy before items are introduced to treatment, with about a 5% chance that the baseline slope is at or below 0. To illustrate this effect, we can look at the estimated probabilities for an individual set. For example, if we take a hypothetical set of words that is introduced to treatment after five baseline probes, the probability of a correct response to items in that hypothetical set increases from approximately 2% at the beginning of the study to 7% before the set is introduced to treatment. The effect of level change was 4.76 (90% CI [3.37, 6.10]), meaning there was a large increase in the probability of a correct response after the introduction of sets to treatment. For example, in the same hypothetical set (introduced to treatment after five baseline probes), the likelihood of a correct response increases from 7% to 90% likelihood directly following introduction of the set to treatment.

The slightly negative slope change ($b = -0.23$; 90% CI [−0.67, 0.22]) was derived by comparing the slope in the treatment phase to the slope in the baseline phase. In general, performance across observations within each phase remained relatively constant (see Figure 3) with a small increase in naming performance during the baseline phase. The negative change in slope, therefore, reflects rapid improvement in naming accuracy on each set after its

Figure 3. Multiple-baseline data depicting naming accuracy over time for participant B.N. Three pretreatment probes for each word set were followed by a 7-week treatment period with weekly probes for naming accuracy and two posttreatment probes. These probes are divided between the baseline phase before the introduction of treatment (B1, B2, B3, B4, . . .) and the treatment phase (T1, T2, T3, . . .) after the introduction of treatment. Additionally, follow-up probes were completed at 3, 6, and 12 months (F1, F2, F3). The dotted line represents when each set of words was introduced to treatment, separating the baseline and treatment phases.

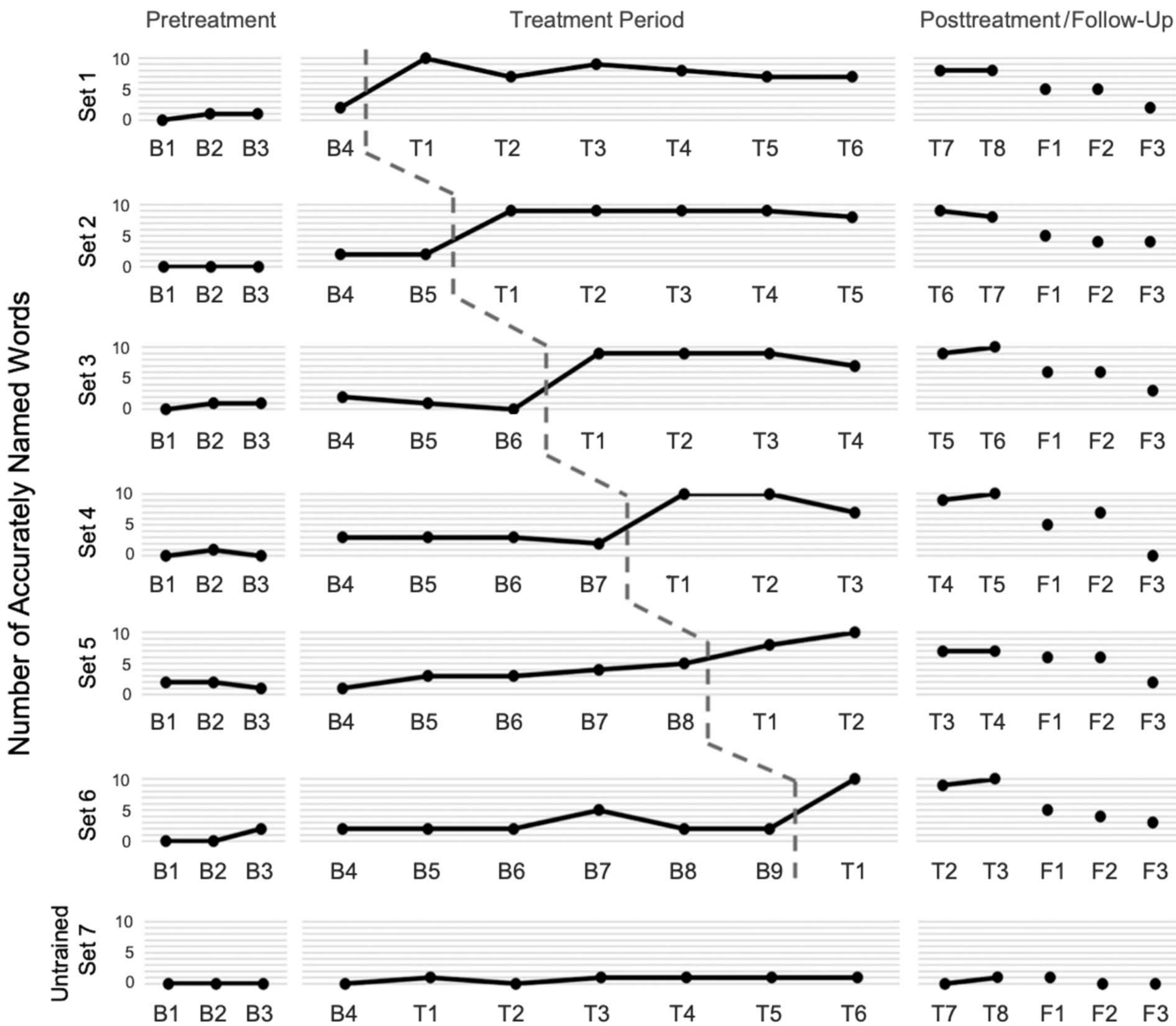


Table 2. Summary of results, represented as logit values, from the interrupted time series model with 90% credible intervals (lower bound at Q5, the fifth percentile, and upper bound at the Q95, the 95th percentile).

Interrupted time series model				
Parameter	**Estimate**	**Estimated error**	**Lower bound Q5**	**Upper bound Q95**
Intercept	–3.70	0.65	–4.85	–2.72
Baseline slope	0.23	0.14	0.00	0.47
Level change	4.76	0.84	3.37	6.10
Slope change	–0.23	0.28	–0.67	0.22

Note. Estimates reflect data from model including both trained and untrained sets.

introduction to treatment, with gains maintained throughout the treatment phase. For example, the likelihood of a correct response for items in a hypothetical set introduced to treatment after five baseline probes changes slightly during the treatment phase from 90% (after the introduction to treatment) to 88% (at the last treatment phase probe).

Investigating Generalization to Untrained Words

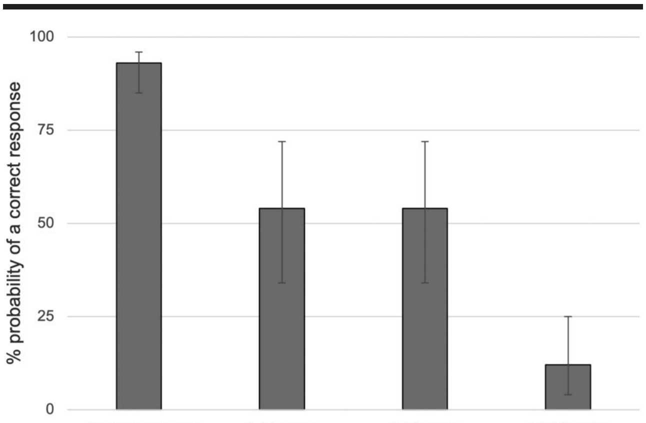
The estimated change in naming accuracy for untrained words was less than one word gained (0.75; 90% CI [0.22, 1.55]) of the possible 10 untrained words. This predicted change for untrained items is consistent with B.N.'s observed performance (i.e., no words named before the initiation of treatment and no more than one word named accurately during probes after the initiation of treatment).

Assessing Maintenance of Trained Items Relative to Posttreatment

The results of the Bayesian generalized mixed-effects analysis of maintenance (see Figure 4) indicated a 93% probability (90% CI [0.86, 0.97]) of a correct response at posttreatment for trained items. This high probability decreased to about a 54% probability (90% CI [0.34, 0.72]) of responding accurately on probes collected at the 3-month follow-up and remained at a 54% probability (90% CI [0.34, 0.72]) for the 6-month follow-up. At 1 year posttreatment, naming performance again decreased, with a 12% (90% CI [0.04, 0.26]) probability of producing a correct response.

Performance on standardized assessments over time. The participant's gradual decline in naming accuracy for trained words over the 12-month follow-up period was observed in the context of an abrupt decline in the use of Anki to practice naming posttreatment as well as gradually decreasing scores on standardized assessments, particularly those assessing semantic knowledge (see Table 1). From pretreatment through the final follow-up, scores on confrontation naming tests/subtests decreased from 10/60 to 2/30 on the Boston Naming Test and from 7.6 to 6.3 on the WAB-R. We also observed decreasing scores on the Pyramids and Palm Trees Test (13/14 to 11/14), the MMSE (27/30 to 23/30), and digits forward (from 6 to 4 digits). Scores on a test of single-word comprehension, the Peabody Picture Vocabulary Test, remained stable throughout the study (score of 8/16 before treatment and 9/16 at 1 year posttreatment).

Figure 4. Percent probability of a correct response on trained items from posttreatment testing through 3-, 6-, and 12-month follow-up visits including 90% confidence intervals.



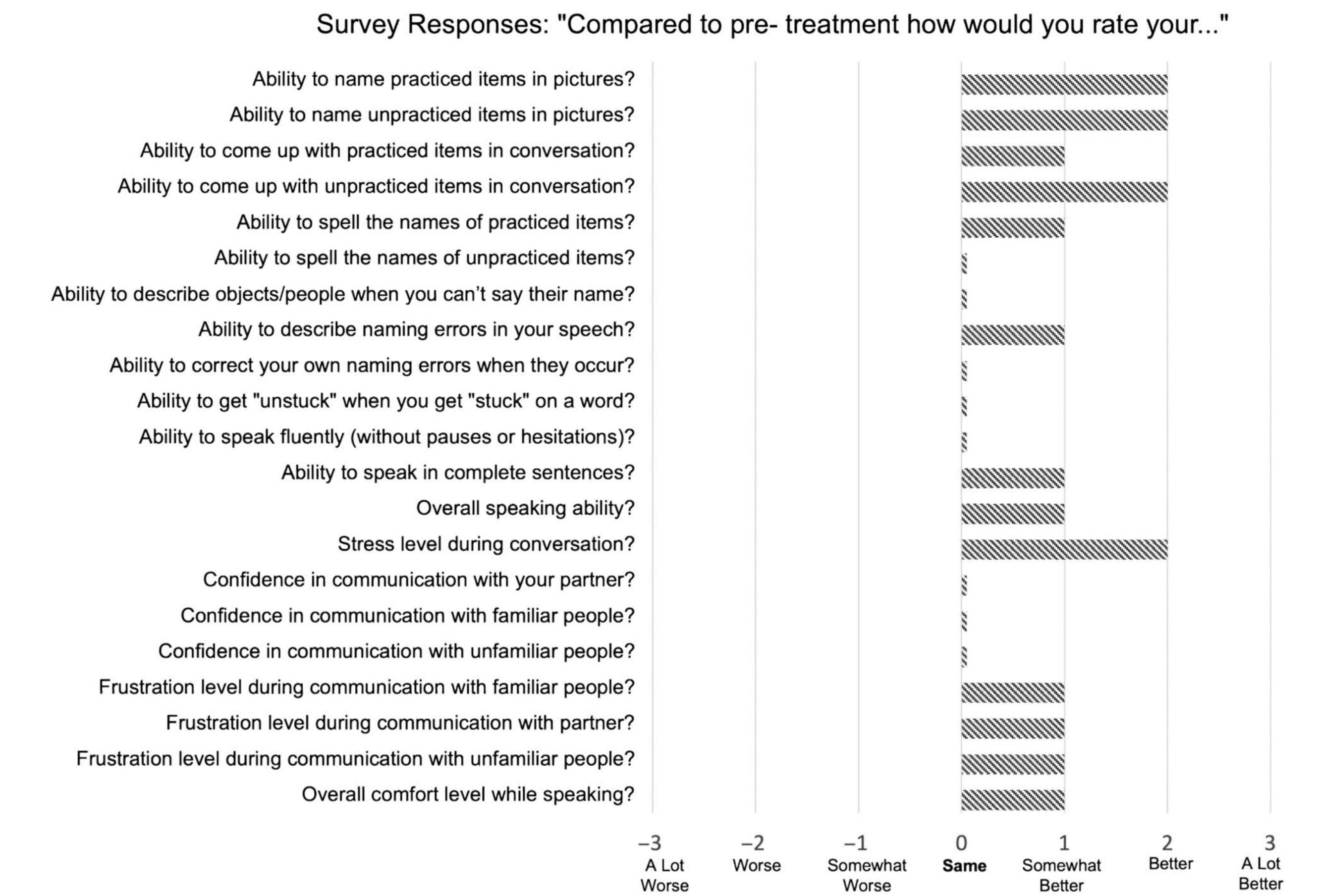
Posttreatment Survey Results: Self-Assessment of Change

On the posttreatment survey (see Figure 5), the participant reported either stability or positive change in response to all questions. B.N. reported that he perceived the greatest change (a rating of "better") in his ability to name practiced and unpracticed items, his ability to recall unpracticed items in conversation, and his level of stress during conversation. He also reported some change (a rating of "somewhat better") in his overall speech and comfort level while speaking, as well as his ability to recall practiced items in conversation, spell practiced items, detect errors in his speech, and speak in complete sentences. In addition, the participant reported that his frustration level when communicating with different communication partners was somewhat better. No change was reported on the nine remaining items, including measures of confidence when communicating.

Discussion

The purpose of this study was to investigate the utility of a self-administered naming treatment. The participant, B.N., presented with svPPA and a history of severe TBI. Following treatment, B.N. showed robust improvements in naming accuracy for trained words, despite reduced daily practice relative to the recommended regimen (30 min per day). For B.N., who completed treatment during an early and relatively mild stage of PPA, an average of 10 min of practice per day was sufficient to relearn each set of 10 words, and he was able to recall those words with a high level of accuracy following treatment. We estimated that the effect size, as a direct result of the self-administered treatment (i.e., accounting for a rise in baseline-phase performance), was 35 out of 60 trained words. B.N.'s success relearning words is commensurate with the positive findings of previous naming treatment studies in svPPA. Specifically, B.N.'s posttreatment

Figure 5. Results of the posttreatment survey, assessing B.N.'s own perception about his performance in treatment as well as the impact of treatment on everyday communication.



performance (about 93% naming accuracy) falls within the average range of results observed in svPPA participants who completed clinician-directed LRT (Henry et al., 2019), indicating that a self-directed Anki treatment was an effective approach for B.N. to relearn personally relevant words. B.N. himself also reported positive effects of naming treatment, specifically, in his ability to recall words during conversation and other communicative abilities (see Figure 5).

The positive results in the current study add to the small pool of single-case experimental design treatment studies and case studies investigating the use of spaced retrieval in naming treatments for svPPA (Bier et al., 2009; Evans et al., 2016; Meyer et al., 2018; Reilly, 2016) as well as self-administered computer-based treatments for svPPA (Evans et al., 2016; Heredia et al., 2009; Krajenbrink et al., 2020; Lavoie et al., 2020; Savage et al., 2014, 2015a). In the current study, we expanded on the positive findings of these previous studies by investigating a treatment that combines strategy training with spaced retrieval training. Additionally, we investigated the effectiveness of treatment for an individual with both svPPA and a previous history of severe TBI.

Treatment for Individuals With Concomitant Conditions

Individuals with complex neurological histories are typically excluded from PPA treatment research (e.g., Schaffer et al., 2022). The participant in the current study was excluded from at least two other treatment studies because of his neurological history. However practical or necessary these exclusionary criteria may be, the result is that little is known about the feasibility and efficacy of speech-language interventions for individuals with PPA and a history of a concomitant neurological disorder such as TBI.

The results of this study indicate that, relative to individuals presenting with svPPA without TBI, people with concomitant neurological diagnoses may benefit from treatment to a comparable degree. Our findings, however, are from a single participant and may not have the same implications for all participants with svPPA and history of TBI, particularly individuals experiencing more severe residual symptoms of brain injury, or those who report different sites of brain injury. A larger study, including individuals who are representative of the heterogeneity of TBI and PPA, would help determine for whom this approach is most suitable.

Practicing speech-language pathologists who work with PPA are likely to encounter individuals with comorbid cognitive-communication disorders including those resulting from TBI, which is observed disproportionately in individuals with FTD (Deutsch et al., 2015; LoBue et al., 2016), as well as comorbidities related to sensory processing. For example, approximately two-thirds of individuals over 70 years of age experience some degree of hearing loss (Goman & Lin, 2016), suggesting that hearing loss frequently co-occurs with other neurogenic disorders affecting older people. As studies of naming treatment continue to grow in number, more can be done to explore the utility of treatment for individuals with comorbid cognitive-communication and sensory disorders. For example, a previous single participant study reported positive effects for an established treatment for nfvPPA (Henry et al., 2019) that was adapted for an individual with concomitant hearing loss through structured modifications to the intervention (Schaffer et al., 2022).

Generalization Effects in the Context of svPPA

As noted in recent reviews of behavioral treatments for PPA (e.g., Cadório et al., 2017; Jokel, 2019), generalization to untrained targets in the context of a neurodegenerative disorder is difficult to achieve, particularly for individuals with svPPA who are experiencing a degradation of semantic memory. Because some clinician-directed treatments that have included training of self-cueing strategies have resulted in increased naming accuracy for untrained items (Beales et al., 2016, 2021; Henry et al., 2013, 2019), we investigated whether we may also see an increase in naming accuracy on untrained items after a self-administered and strategy-based approach. Given his relatively mild semantic deficits, we anticipated that B.N. might draw on residual semantic and word-form knowledge to self-cue for untrained words. However, given the previous mixed findings for individuals with svPPA, particularly in similar self-administered treatment studies (Evans et al., 2016; Heredia et al., 2009; Krajenbrink et al., 2020; Lavoie et al., 2020; Savage et al., 2014, 2015a), we did not set an a priori hypothesis regarding generalization to untrained words. Despite B.N.'s relatively spared semantic memory, we did not observe a meaningful increase in accuracy for untrained targets and no improvement was present on a test of confrontation naming.

An additional factor that may have contributed to the lack of generalization in this case is the mode of treatment delivery. Unlike in previous studies targeting strategic self-cueing that did show generalization effects (Dial et al., 2019; Henry et al., 2013, 2019), B.N. did not have the benefit of a clinician explicitly describing, reinforcing, and encouraging self-cueing strategies in real time throughout the course of treatment, which may have affected his ability to generalize word-finding strategies. In other studies of self-administered computer-based naming treatment for svPPA, some researchers have demonstrated generalization to untrained exemplars for trained words or untrained contexts (Heredia et al., 2009; Lavoie et al., 2020; Savage et al., 2014), but only one case, reported by Lavoie and colleagues, has shown significant improvements on untrained words. Despite the inclusion of written prompts meant to encourage strategy use, our findings were consistent with the majority of previous studies investigating self-administered computer-based interventions.

Maintenance of Treatment Gains and the Impact of Participant Motivation

We hypothesized that, with ongoing practice of trained words, B.N. would maintain gains through 12 months, consistent with participants with svPPA who have participated in clinician-directed treatment targeting strategic self-cueing (Henry et al., 2019). However, the observed results were more consistent with reports of maintenance from other computer-based self-administered treatments (Heredia et al., 2009; Lavoie et al., 2020; Savage et al., 2015a), which describe maintenance up to 6 months with some decrease in accuracy during the follow-up period. B.N.'s ability to name trained items decreased over the year following the formal training period, with an estimated 4% probability of correct response at pretreatment (see Table 2), 93% at posttreatment, 54% at 6 months posttreatment, and 12% at 12 months posttreatment (see Figure 5). Despite a gradual decrease in performance, some maintenance of gains compared to pretreatment was demonstrated through 6 months. Likelihood of a correct response at the 12-month follow-up also appears to be slightly higher than at pretreatment. Notably, this partial maintenance of gains in naming of trained words occurred in the context of infrequent practice as well as decreasing cognitive and language abilities (e.g., poorer confrontation naming, lower MMSE score).

Maintenance effects may be impacted by an individual's own motivation to continue practicing words. In this study, the participant was diligent in practicing during the treatment period, but his motivation appeared to decrease after treatment. He practiced only 6–7 times between each of the follow-up observations. An individual's motivation to participate in treatment is an important consideration in treatment of PPA (Taylor-Rubin et al., 2019) and is of particular importance when considering self-administered treatment options. Increased clinician involvement may help to maintain motivation by ensuring that a patient or care partner is able to introduce personally relevant and appropriately challenging practice items over the course of a self-administered treatment. Accountability in the form of check-in appointments with a clinician, where possible, may also help to encourage periods of increased independent practice (i.e., boosters), which have been shown to support maintenance of gains for individuals with svPPA. Savage et al. (2015a) found that such boosters helped maintain treatment gains if participants began showing a decline in naming accuracy. For a self-administered treatment using Anki, a clinician could conceivably track a patient's independent use of Anki, then intervene if either adherence or performance showed decline.

Several factors may have contributed to B.N.'s reduced practice after treatment including the absence of new words being introduced into treatment, decreased accountability as he was no longer asked to regularly meet with the clinician, and gradual decline in cognitive–linguistic function. As cognitive and linguistic abilities decline due to svPPA, an individual's awareness of their own deficits and ability to appraise their performance may also diminish (Savage et al., 2015b), limiting the utility of self-directed treatment approaches. A lack of insight into deficits is particularly important to consider and evaluate for individuals with a concomitant history of TBI, as anosognosia is also a common symptom of moderate-to-severe TBI (Steward & Kretzmer, 2022). However, clinician-guided treatment can be fruitful even in more advanced cases, providing support to individuals with svPPA and their families through tailored and dynamic treatment plans that evolve with progression. Potential approaches include compensatory tools, such as a communication wallet with pictures and words related to topics of everyday conversation (Fried-Oken et al., 2010; Rogalski & Khayum, 2018) and communication partner training (Volkmer et al., 2021).

Strengths, Weaknesses, and Directions for Future Research

The method of delivering treatment used in this study was modeled in part on a case reported by Evans et al. (2016), in which one individual with svPPA regularly practiced words using Anki over a 20-month period. While the current study investigates a smaller training set (60 words compared to 591), we were able to include experimental controls that were not present in the Evans et al. study, including the use of a multiple-baseline design as well as an untrained set of personally meaningful words that was matched with the trained word sets on a variety of relevant linguistic parameters. The current study, therefore, reinforces and extends the conclusions of the Evans et al. (2016) study, demonstrating the utility of self-administered computer-based approach using Anki to deliver treatment.

One weakness of the current study is that we did not investigate generalization of word retrieval beyond the word level to connected speech. While the ultimate objective in any naming treatment is that the person with PPA improves their ability to communicate in conversation, the potential for generalized improvements in word retrieval during connected speech tasks following naming treatment has not been well studied, and thus far, findings with regard to this type of generalization are mixed (Beales et al., 2018). Further research is needed to investigate generalization to connected speech for naming treatments in PPA, including self-administered treatment options.

Thus far, investigation of self-administered computer-based naming treatment in svPPA has included only single-case experimental studies (e.g., Evans et al., 2016). In the future, research should investigate treatment effects with a larger group of participants with svPPA to better determine the benefits of self-administered treatment. Specifically, group-level analysis would allow for a statistical comparison of treatment effects between a self-administered naming treatment and a comparable, clinician-led naming treatment. The results of such a comparison would provide valuable information about the relative benefit of these different treatment approaches that could be used in clinical decision making. Such studies could also investigate questions about the impact of item saliency on learning, a crucial aspect of treatment for individuals with svPPA, who may struggle to recognize items that are not relevant to their daily lives (i.e., lower frequency and less familiar words; Gorno-Tempini et al., 2011; Hardy et al., 2023).

For group studies and single-case studies alike, exploring the potential prophylactic benefits of self-administered treatment for individuals with svPPA is of particular importance. The number of words that an individual with svPPA can expect to relearn from treatment is limited even when treatment spans more than 1 year (e.g., Evans et al., 2016). Therefore, future studies may choose to focus self-administered treatments on a core set of personally tailored, functional words. A prophylactic approach to treatment, endorsed in several naming treatment studies (e.g., Meyer et al., 2018; Reilly, 2016), has the potential benefit of preserving the most functional words in order to extend the use of verbal communication for activities of daily living and personally salient topics. For example, Reilly (2016) utilized a combination of spaced retrieval and error-reduced learning to train a large set of words and found that trained words were retained for longer compared to untrained words. These trained words could also be incorporated into assistive and augmentative communication tools, such as communication boards to maintain communication in the face of declining spoken language.

Future research should continue to explore the effects of various intervention approaches in populations presenting with disorders that co-occur with PPA, such as TBI. Because people with complicated medical histories and co-occurring disorders are often excluded from large-scale clinical trials, there may be unique considerations for those individuals that have not yet been addressed in the literature. Single-case experimental design studies are well suited for exploring the relationship between specific cognitive–linguistic profiles and the effects of treatment (Best et al., 2019). Such studies, therefore, are an important tool to inform appropriate clinical care for individuals who do not fit well into specific diagnostic categories or who have additional considerations beyond their primary diagnosis.

With respect to the statistical analyses in this study, we employed Bayesian generalized mixed-effects modeling, including an interrupted time series model (Huitema & Mckean, 2000), a relatively novel method for analyzing multiple-baseline data in the field of aphasiology. The primary strength of this approach is that we were able to account for the magnitude of change and variability in slope during the baseline phase. In accounting for change during the baseline phase, the estimated treatment effect may be more conservative than an effect size calculated from pre- and posttreatment performance alone. However, this conservative estimate of the causal effect of intervention is aligned well with the theoretical rationale for the multiple-baseline, single-case experimental design. One possible drawback of our statistical approach is that it is more complex than other methods of analyzing single-case experimental data. For those considering a similar approach, the advantages and disadvantages of using Bayesian interrupted time series models have been further explored in a tutorial by Cavanaugh et al. (2023).

Clinical Implications

By increasing the availability of self-directed treatments for individuals with svPPA, we can expand the number of people who are able to participate in speech-language intervention. Self-directed treatment programs also provide clinicians with a tool they can recommend for clients to continue practicing words after they are discharged from speech-language treatment or to bridge the gap between periods of formal, clinician-directed treatment. This is particularly important for individuals who are restricted in the amount of treatment they are able to receive due to limited coverage from third-party reimbursers. Self-directed treatments, along with increasing use of remote treatment, may also help to overcome a variety of barriers faced by those seeking speech-language intervention for svPPA, such as difficulty finding speech-language pathologists with appropriate expertise in one's geographic area.

In many ways, Anki and similar self-directed programs are ideally suited for individuals in the early stages of svPPA. A person with mild svPPA or their partner can keep an ongoing list of personally relevant words they notice are difficult to recall. Subsequently, small groups of functional but challenging words can then be added regularly to the Anki database and practiced as part of a daily routine. The spaced retrieval algorithm assures that the most challenging words are practiced most frequently and that, as words become more challenging in the face of degrading semantic knowledge, they can be maintained through repeated practice.

Optimal dosage for the use of Anki as a self-directed naming treatment for svPPA is an open question. Guidance for PPA treatment generally, both self-administered and clinician led, is lacking, and reported dosage varies widely among treatment studies (see Wauters et al., 2023). One study of individuals with stroke-induced aphasia suggests that, due to the adaptive nature of the Anki program, participants needed progressively less practice over time to retain word learning (Quique et al., 2022). However, in the face of declining cognitive–linguistic abilities, as is the case in svPPA, retention of words may require more sustained practice. Increased clinician involvement and periodic introduction of novel practice items may help to maintain motivation and consistent practice (Savage et al., 2015a). Self-directed use of Anki or similar programs may be less suited to those who are not able to monitor their own naming difficulty, those who are not motivated to participate in regular practice, and those who are in later stages of PPA and therefore may not have the cognitive skills to operate the program independently.

Conclusions

A growing body of evidence shows positive effects of intervention in PPA (e.g., Cadório et al., 2017; Carthery-Goulart et al., 2013; Cotelli et al., 2020; Volkmer, Rogalski, et al., 2020). Nevertheless, few studies have investigated methods for reducing barriers to service options, such as the implementation of teletherapy (e.g., Dial et al., 2019) and self-administered treatments (e.g., Evans et al., 2016). Additionally, prior studies have excluded individuals reporting concomitant neurological conditions, including TBI. To address these gaps in the literature, we (a) implemented a treatment designed to maximize learning (via spaced retrieval training) as well as the use of self-cueing strategies, (b) investigated the effects of naming treatment for an individual with both svPPA and history of severe TBI, and (c) expanded on previous work reporting the benefits of self-directed naming treatment for svPPA. We documented significant gains in naming accuracy, which decreased gradually over the year following treatment. Compared to pretreatment, gains were partially maintained though at least 6 months posttreatment despite infrequent practice beyond the treatment period. These results are complemented by the participant's self-perceived improvements in naming ability and decreased stress/frustration level during conversation. Taken together, these findings indicate that self-directed treatments represent a beneficial resource for people with mild-to-moderate svPPA who either have limited access to or are looking to supplement clinician-directed intervention. Future studies should explore benefits of self-directed naming treatment in comparison to clinician-directed naming treatment as well as the potential prophylactic benefits of self-directed naming treatments. With a more fully developed evidence base for self-directed treatments, families affected by PPA and their care providers will be better equipped to make decisions regarding speech-language interventions.

Author Contributions

Gary Robinaugh: Conceptualization (Supporting), Data curation (Supporting), Formal analysis (Lead), Methodology (Lead), Project administration (Lead), Software (Lead), Visualization (Lead), Writing – original draft (Lead), Writing – review & editing (Lead). **Stephanie M. Grasso:** Conceptualization (Lead), Data curation (Lead), Formal analysis (Supporting), Methodology (Supporting), Project administration (Supporting), Supervision (Lead), Visualization (Supporting), Writing – original draft (Supporting), Writing – review & editing (Supporting). **Maya L. Henry:** Conceptualization (Supporting), Formal analysis (Supporting), Funding acquisition (Lead), Methodology (Supporting), Resources (Lead), Visualization (Supporting), Writing – original draft (Supporting), Writing – review & editing (Supporting). **Robert Cavanaugh:** Formal analysis (Supporting), Methodology (Supporting), Software (Supporting), Visualization (Supporting), Writing – original draft (Supporting), Writing – review & editing (Supporting).

Data Availability Statement

The data set generated and analyzed during the current study is available in the Open Science Framework repository (https://osf.io/drfv5/?view_only=d3bf293b1b0f44f1a9fc06ee2747c745).

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Appendix A

Bayesian Generalized Mixed-Effects Model: Naming Performance

To evaluate treatment effects, we used an interrupted time series approach to analyze single-case, multiple-baseline design data, as described by Huitema and Mckean (2000), Moeyaert et al. (2017), and Cavanaugh et al. (2023) using the BRMS package (Bürkner, 2017) in R. We employed the models described by Cavanaugh et al. modeling item-level responses (correct or incorrect) with population-level effects (i.e., fixed effects) of intercept, baseline slope, level change, and slope change. Models included group-level intercepts for items nested within sets with corresponding group-level slopes for the primary three population-level effects, allowing each item and set to have their own intercept (an estimate of performance prior to the first baseline session), baseline slope, level change, and slope change.

While data from Sets 1–6 were used to estimate the baseline slope, level change, and slope change, data from Set 7 were only included in the estimate of the baseline slope and did not influence the estimate of level change or slope change. Because group-level effects permitted each list to have its own intercept, baseline slope, level change, and slope change, this approach allowed us to include data from Set 7 to better estimate the baseline slope while obtaining separate treatment effect estimates for trained items (Sets 1–6) and untrained items (Set 7).

Prior distributions were as follows, in logits. The prior distribution on the intercept was a normal distribution with an *M* of −1 and an *SD* of 2. This prior reflects our a priori knowledge of the stimuli selection procedure, such that performance at the start of baseline should be roughly 30% correct. Priors on the population-level effects were consistent of a normal distribution with an *M* of 0 and an *SD* of 2. This prior makes no assumptions about the direction of the treatment effects but permits the possibility of relatively large treatment effects such as those found in the study of Quique et al. (2022). A relatively wide prior on the group-level effects (Normal, 0, 2) was intended only to guide initial sampling to plausible values. The model also includes a correlation parameter to describe the correlation between the group-level intercept and the group-level slope. A Lewandowski–Kurowicka–Joe (LKJ) prior with LKJ = 2 was used for the correlation parameter such that extreme correlation values would be less likely.

We estimated the posterior distributions for our interrupted time series model using Markov Chain Monte Carlo simulation with four chains and 4,000 total samples per chain. To account for a calibration period, 1,000 samples per chain were discarded and not included in subsequent analysis. Posterior predictive checks confirmed the models adequately fit the data. The estimated split-half potential scale reduction factor values were less than 1.01, with effective sample sizes exceeding 1,000 for all parameters.

To examine whether our choice of priors had undue influence on results, we completed a sensitivity analysis of the model, comparing the results of the same model given variations in the priors (see Supplemental Material S2). Changes to the prior distribution resulted in only minor changes to posterior estimates, no changes to the overall interpretation of the model results, and minimal impact on the overall effect size estimates. Additional details including R code can be found in Supplemental Material S1.

Appendix B

Bayesian Generalized Mixed-Effects Model: Maintenance of Trained Words

Maintenance of gains for trained words was evaluated using a Bayesian generalized mixed-effects model with the BRMS package (Bürkner, 2017) in R. This model included a four-level categorical population-level effect of time point (posttreatment, 3-, 6-, and 12-month follow-ups). Posttreatment was used as the reference value such that effects should be interpreted relative to posttreatment performance. Words were nested into sets and included as a group-level effect to reflect the structure of the data set, which includes six sets of trained words introduced to treatment at different time points during treatment. A summary of both the population- and group-level effects can be found in Supplemental Material S1.

We used weakly informative priors in the model including a normal distribution with an *M* of 0 and an *SD* of 2 for both population- and group-level effects. Similar to the interrupted time series model described in Appendix A, these priors do not make any assumptions about the direction of change in performance comparing follow-up time points to posttreatment. A normal distribution with an *M* of −1 and standard an *SD* of 2 was used as the prior distribution of the intercept.

We estimated the posterior distributions for our interrupted time series model using Markov Chain Monte Carlo simulation with four chains and 5,000 total samples per chain. To account for a calibration period, 1,000 samples per chain were discarded and not included in subsequent analysis. We then confirmed that the models adequately fit the data using posterior predictive checks. The estimated split-half potential scale reduction factor values were less than 1.01, with effective sample sizes exceeding 1,000 for all parameters.

To examine whether our choice of priors had undue influence on results, we completed a sensitivity analysis of the model, comparing the results of the same model given variations in the priors (see Supplemental Material S2). Changes to the prior distribution resulted in only minor changes to posterior estimates and no changes to the overall interpretation of the model results. Additional details including R code can be found in Supplemental Material S1.