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Exploring the Efficacy of Teletherapy in Individuals with Primary Progressive Aphasia

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Exploring the Efficacy of Teletherapy in Individuals with Primary Progressive Aphasia

by

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Dedication

In loving memory of Mr. D. Though our time together was short, you were an important part of this project. I was honored to have been a small part of your journey and to work with you and your family. It is because of individuals like you that we at the Aphasia Research and Treatment Lab are passionate about working with individuals with primary progressive aphasia and their families. Your humor during our sessions and your positivity were sources of encouragement for me. Thank you.

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Abstract

Exploring the Efficacy of Teletherapy in Individuals with Primary Progressive Aphasia

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There is growing evidence suggesting that restitutive treatment for speech and language deficits may have substantial, lasting benefits for individuals with primary progressive aphasia (PPA). Nonetheless, reimbursement restrictions, geographical constraints, and other factors limit the ability of clinicians to reach individuals with this diagnosis. The advent of teletherapy has allowed clinicians to treat otherwise unreachable patients and to increase treatment dosage for other patients. Currently, however, there is limited data regarding the benefit of teletherapy in PPA.

We present data from two treatment studies in PPA, comparing treatment outcomes for face-to-face interventions with teletherapy. Specifically, we examined Lexical Retrieval Training (LRT) in fluent patients and Video Implemented Script Training in Aphasia (VISTA; a treatment for speech production/fluency) in nonfluent/agrammatic patients. Interventions were implemented with 25 participants, with 9 participants receiving teletherapy. We compare data from primary outcome

measures as well as from generalization measures at post-treatment and three and six months post-treatment.

For LRT, participants showed significant improvement on trained and untrained nouns, as well as the *Boston Naming Test* (*BNT*) at post-treatment. Maintenance of gains was observed for trained and untrained nouns at three and six months post-treatment and for the *BNT* at 3 months post-treatment. A direct comparison of traditional and teletherapy sub-groups revealed no differences between the two groups at any time point for any measure.

For VISTA, participants showed significant improvement on trained and untrained scripts, as well as the *Northwestern Anagram Test* (*NAT*) at post-treatment. Maintenance of gains was observed for trained scripts and the *NAT* at three and six months post-treatment and for untrained scripts at 3 months post-treatment. A direct comparison of traditional and teletherapy sub-groups revealed no differences between the two groups at any time point for any measure.

These results lend additional evidence to current research that documents positive treatment outcomes in individuals with PPA. In addition, these treatment benefits are lasting, both for trained and untrained targets. Finally, we demonstrate that treatments administered via face-to-face sessions and those administered via teletherapy produce comparable results at post-treatment and follow-up. Thus, teletherapy is a viable approach for treatment delivery in PPA.

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1. Introduction

Primary progressive aphasia (PPA) is a degenerative neurological condition in which speech and language functions progressively deteriorate in the context of relatively spared cognitive functioning. Individuals with PPA are less likely to be referred for or offered behavioral treatment for speech-language deficits due to negative expectations concerning the efficacy of behavioral treatment in neurodegenerative disease, as well as a lack of understanding of the disorder by many clinicians (Henry et al., 2013a). However, there exists a growing body of research characterizing the disorder and various treatment approaches in PPA. Treatment research has focused on various aspects of disease management, including restitutive treatments to improve speech-language functions, treatments to address life participation and activity limitations, and augmentative/alternative approaches to maintain communication throughout the progression of the disease. Research on restitutive treatments in PPA has established modest evidence supporting the utility of restitutive speech language treatment in PPA, with most studies focusing on lexical retrieval deficits (for review, see Croot, Nickels, Laurence, & Manning, 2009; Jokel, Graham, Rochon, & Leonard, 2014). Most studies demonstrate gains on trained and, to some extent, untrained language measures, with treatment appearing to slow progression of language deficits on trained language skills (Jokel, Rochon, & Leonard, 2006; Farrajota et al., 2012).

Although restitutive treatment has been established as feasible and efficacious in individuals with PPA, questions of access to services remain. Most individuals do not live near rehabilitation centers that offer specialized treatment for PPA, and thus they do not receive services to address their linguistic deficits. Recent advances in technology have resulted in a substantial increase in the use of live videoconferencing, known as "teletherapy," to treat speech and language disorders. Teletherapy offers the advantages of increased access to speech-language services and reduced cost of treatment, and to date has shown promising outcomes in the management of chronic aphasia, dysarthria, voice disorders, and Alzheimer's dementia (for review, see Cherney & Van Vuuren, 2012; Edwards, Stredler-Brown, & Houston, 2012; Hall, Boisvert, & Steele, 2013; Jelcic et al., 2014). As video and internet technologies continue to improve and barriers related to licensure and reimbursement are resolved, teletherapy will play a greater role in the management of chronic conditions.

In this study, we examined the potential benefit of language intervention delivered via teletherapy to individuals with PPA. In an effort to explore the utility of teletherapy for individuals with PPA, treatment outcomes in teletherapy were compared to outcomes in traditional therapy in a group of participants with PPA to determine whether comparable gains are achieved via teletherapy.

1.1 Primary Progressive Aphasia (PPA) and its Variants

Primary Progressive Aphasia (PPA) is a progressive neurological condition that results in a relatively isolated impairment of language, and is caused by neurodegenerative disease that affects areas of the brain that support communication. Although other cognitive functions may be affected in the later stages of PPA, speech and language are the most prominent deficits in the initial stages of the disease, and they remain the most impaired domains throughout disease progression (Gorno-Tempini et al., 2011). Thus, activities of daily living are generally maintained with the exception of those related to language (e.g. speaking on the telephone) (Gorno-Tempini et al., 2011).

There are three recognized variants of PPA, each presenting with a distinct set of speech and language characteristics and pathologies. The non-fluent/agrammatic variant of PPA (nfvPPA) presents with non-fluent spontaneous speech, agrammatism, and deficits in motor speech (Gorno-Tempini et al., 2011; Henry et al., 2013a). Atrophy is prominent in the left posterior and inferior frontal lobe, which is responsible for grammar and speech production (Gorno-Tempini et al., 2011; Wilson et al., 2010). The semantic variant of PPA (svPPA) is characterized by impaired single-word comprehension, object knowledge, and naming due to an impaired semantic system. This variant is associated with bilateral atrophy in the anterior temporal lobe, with atrophy typically greater in the left than the right hemisphere (Gorno-Tempini et al. 2011; Henry et al., 2013b; Hodges & Patterson, 2007). Lastly, the logopenic variant (lvPPA) presents with phonological deficits, which manifest clinically as impairments in naming, repetition, and comprehension. In lvPPA, atrophy is present in the left temporoparietal junction, including the left posterior superior and middle temporal gyri and inferior parietal lobule. This region is responsible for phonological processes involved in both the comprehension and production of language (Gorno-Tempini et al., 2008; Henry & Gorno-Tempini, 2010).

Impaired lexical retrieval results in difficulty retrieving or producing the names of objects due to a breakdown in one or more levels (semantic, phonological, and/or motor planning) in the complex process of word retrieval (Mayer & Raymer, 2004). Deficits in lexical retrieval are present in all three variants of PPA, although to differing degrees. Whereas lexical retrieval impairment is an early and prominent characteristic in svPPA and lvPPA, word retrieval abilities are spared in nfvPPA until the advanced stages of the disease (Gorno-Tempini et al., 2004). In svPPA, deficits in lexical retrieval are attributed to degraded semantic knowledge that is thought to weaken access to intact phonological representations (Lambon Ralph, McClelland, Patterson, Galton, & Hodges, 2001). Conversely, lvPPA patients present with relatively preserved semantic knowledge, with lexical retrieval deficits likely resulting from phonological storage and assembly deficits, as evidenced by phonological paraphasias commonly present in speech (Gorno-Tempini et al., 2008; Wilson et al., 2010). Thus, the majority of treatment studies in svPPA and lvPPA have focused on lexical retrieval deficits. Treatment studies in nfvPPA have also focused on lexical retrieval deficits but to a lesser degree, given that other features, such as sentence production and apraxia of speech, are the central and defining deficits.

1.2 Speech and Language Treatments in PPA

As research has continued to define the characteristics and underlying pathologies of the variants, a modest body of research literature regarding treatment outcomes in PPA has also arisen. The majority of this work has examined lexical retrieval in svPPA, with few studies dedicated to lvPPA and nfvPPA variants. In svPPA, these studies have typically implemented a treatment protocol in which a picture was paired and rehearsed with the written word to activate both semantic and orthographic/phonological representations (Graham, Patterson, Pratt, & Hodges, 1999, 2001; Heredia, Sage, Lambon Ralph, & Berthier, 2009; Jokel et al., 2006; Mayberry, Sage, Ehsan, & Lambon Ralph, 2011; Savage, Ballard, Piguet, & Hodges, 2013; Savage, Piguet, & Hodges, 2015; Snowden & Neary, 2002). However, while these studies resulted in significantly improved naming performance for trained items, the majority of these gains were item- and context-specific, and only a few studies demonstrated maintenance up to 6 months post-treatment (Heredia et al., 2009; Jokel et al., 2006; Savage et al., 2015). The item- and context- specific gains demonstrated in these studies suggest that new learning and lexical retrieval may become dependent on episodic memory and rote memorization as semantic representations degrade. Other studies have implemented elaborate cueing hierarchies that utilize semantic, phonological, and orthographic information to support retrieval (Bier et al., 2009; Dressel et al., 2010; Henry et al., 2013b; Jokel & Anderson, 2012; Jokel, Rochon, & Anderson, 2010; Newhart et al., 2009). These studies also report improved naming performance, along with generalization to untrained items (Henry et al. 2013b; Jokel & Anderson, 2012; Jokel et al., 2010) and maintenance of gains for two or more months following treatment (Dressel et al., 2010; Henry et al. 2013b; Jokel & Anderson, 2012; Jokel et al., 2010). Thus, a richer variety of cueing modalities may be more beneficial for patients with svPPA, with both semantic and phonological cueing having therapeutic value (Dressel et al., 2010).

While studies on lexical retrieval treatment in lvPPA are fewer, results are promising. Both Newhart et al. (2009) and Henry et al. (2013b) utilized a cueing hierarchy with an lvPPA patient in addition to their svPPA patient, which resulted in improved naming on both trained and untrained items. In addition, the hierarchy implemented by Henry et al. (2013b) sought to encourage self-cueing to promote generalization to other contexts, with both participants reporting improved lexical retrieval and increased confidence in conversation. Another study implemented semantic elaboration training and generative naming in a brief but intensive protocol (Beeson et al., 2011). Treatment effects in naming were significant for trained and untrained sets, and maintained for trained items at six months post-treatment. Furthermore, functional MRI confirmed changes in brain activation patterns that suggested increased activation of preserved regions of the language network following treatment, as well as recruitment of frontal cortical regions. In another study, Croot et al. (2014) implemented a repetition and reading task paired with pictures that was self-administered by an individual diagnosed with lvPPA. Significant treatment effects were observed on trained items but no generalization to untrained items or connected speech was found. Lastly, Trebbastoni, Raccah, De Lena, Zangen, and Inghilleri (2013) applied an experimental repetitive deep transcranial magnetic stimulation (rTMS) treatment. Following sham or real stimulation, the participant underwent a series of cognitive and linguistic tasks to examine whether stimulation led to gains on lexical retrieval in verbal fluency and written narrative tasks. Following real stimulation, a significant increase in phonemic verbal fluency and a significant reduction in the number of syntactic and semantic errors written by the patient were found as compared to both baseline and sham conditions. While only an exploratory study of a novel treatment, results provide preliminary evidence for using rTMS to enhance plasticity within the language network.

Individuals with nfvPPA have also received limited attention in the treatment literature. The few treatment studies available have focused on a range of linguistic deficits associated with the disorder, such as verb and sentence production (Schneider, Thompson, & Luring, 1996), apraxia of speech (Henry et al. 2013a), phonological processing (Louis et al., 2001), and lexical retrieval (Croot et al. 2014; Jokel, Cupit, Rochon, & Leonard, 2009; Marcotte & Ansaldo, 2010). In working with verb and sentence production, Schneider et al. (1996) trained a participant with nfvPPA to produce four transitive verbs in future and past tenses in an unchanging sentence structure with varying vocabulary. Results indicated significant improvement of trained verbs in future and past tenses, with generalization to untrained verbs. Henry et al. (2013a) explored the efficacy of a protocol for facilitating multisyllabic word production that involved structured oral reading of text paired with training to self-detect major speech errors in an individual with apraxia of speech (AOS) associated with nfvPPA. Following treatment, multisyllabic word production in trained text was error-free while major speech errors in untrained text decreased significantly and successfully self-corrected errors increased. Louis et al. (2001) examined a phonological processing intervention in which three individuals with nfvPPA participated in daily sessions targeting segmentation and discrimination of syllables and phonemes. Significant treatment gains were observed on trained tasks in all 3 patients, with a reduction in phonemic paraphasias and improvement on parts of the *Boston Diagnostic Aphasia Examination* (*BDAE*; Goodglass, & Kaplan, 1983) for some participants. As in svPPA treatment research, studies on lexical retrieval in nfvPPA have utilized a protocol in which a picture was paired and rehearsed with the written word (Jokel et al., 2009; Croot et al., 2014). In both studies, significant treatment effects were observed on treated items but did not generalize to untreated items, with maintenance at one month observed in only one participant (Jokel et al., 2009). Marcotte and Ansaldo (2010) also observed significant treatment gains on treated items following a semantic feature analysis intervention, but did not measure for maintenance of gains following treatment. The study also reported changes in brain activation patterns post-treatment, with more extensive activation within typical language networks and recruitment of temporal lobe areas implicated in naming tasks, indicating reactivation and reorganization of the language network following treatment.

Across variants and treatment designs, behavioral treatment of speech-language deficits has been shown to produce immediate gains in all studies undertaken. However, more research is needed to further address deficits other than lexical retrieval difficulties. Research concerning lexical retrieval show that studies that reported generalization to untreated items and/or maintenance of treatment effects typically employed both semantic and phonological approaches to engage multiple language processing components (Beeson et al., 2011; Dressel et al., 2010; Henry et al., 2013b; Jokel & Anderson, 2012; Jokel et al., 2010; Newhart et al., 2009). Furthermore, studies that examined changes in fMRI data from pre- to post-treatment have shown treatment-induced changes in activation patterns, typically resulting in a strengthening of the preserved language network and some recruitment of perilesional left and homologuous right hemisphere areas (Beeson et al., 2011; Dressel et al. 2010; Marcotte & Ansaldo, 2010).

1.3 Special Considerations in PPA that Motivate the Need for Teletherapy

In the last 10 years, teletherapy, or the delivery of intervention and care via remote means, has increasingly been pursued as a viable option in managing adult neurogenic language disorders (Edwards et al., 2012; Hall et al., 2013). Interest in the use of new audio and video technologies for remote management of chronic conditions has been driven primarily by the promise of improved access to medical care at a reduced cost. Furthermore, teletherapy has been endorsed by the American Speech-Language-Hearing Association (ASHA) as a tool to overcome the barriers of access to treatment caused by distance, unavailability of specialists, and impaired mobility (American Speech-Language-Hearing Association [ASHA], 2005).

Individuals with PPA could particularly benefit from teletherapy services. Compared to individuals with stroke-related aphasia, individuals with PPA are under-referred for speech-language pathology services due to lack of knowledge of the disorder and negative assumptions concerning the value of behavioral treatment. Furthermore, in the cases in which a referral is made, speech language pathologists (SLPs) often report feeling unprepared and insecure in providing treatment (Taylor, Kingma, Croot, & Nickels, 2009). Although rehabilitation centers that specialize in the treatment of PPA are available, many individuals typically do not live near these specialized centers. Thus, individuals often do not receive services addressing their communication needs or they must choose to travel unreasonable distances for treatment. Additional considerations include caregiver burdens in neurodegenerative disease, as these differ from burdens encountered by caregivers of non-progressive neurological disorders such as stroke. While mobility in stroke patients may be compromised, caregivers for individuals with PPA must adapt to the constantly changing cognitive and physical status of their patients. As the disease progresses, cognitive functions other than language become implicated as well as motor systems, both of which affect the ease of travel and moving from appointment to appointment. Anxiety and distress may also become an issue as these individuals become increasingly context-dependent, making new, unfamiliar environments challenging to navigate (Rutherford, 2014). Teletherapy offers an avenue of treatment that mitigates travel issues and access to specialists, and allows the individual to access services from the comfort of their home.

1.4 Current Status of Teletherapy in PPA and Stroke-related Aphasia

To date little research has been conducted addressing the feasibility and efficacy of teletherapy in individuals with PPA. Meyer, Getz, Brennan, Hu, and Friedman (2015) examined the benefits of teletherapy in delivering a lexical retrieval protocol within the three types of PPA as compared to traditional therapy. The protocol included a phonological condition, in which an auditorily-presented word was paired with a picture and repeated by the participant, and an orthographic condition, in which the written word was paired with a picture and read aloud and transcribed by the participant. The protocol included both known and unknown items, and at the end of six months of treatment, gains between the teletherapy participants and traditional participants were compared.

The three participants who received teletherapy demonstrated positive treatment effects with both the phonological and the orthographic conditions resulting in significantly greater post-treatment naming accuracy compared to the untrained condition. Each participant was then compared to a small group of individuals with the same variant of PPA that had received the same treatment protocol in person. In each of the three cases, the teletherapy participants showed treatment effects that were within the expected range or larger than those exhibited by the traditional participants. Meyer and colleagues concluded that teletherapy was feasible and effective in providing treatment to individuals with PPA.

Other than Meyer et al. (2015), no study has sought to directly study the benefits of teletherapy as compared to traditional therapy. However, of the two participants treated in Henry et al. (2013b), one participant, “LV,” was treated via teletherapy due to travel constraints. LV underwent a six week lexical retrieval protocol that incorporated a self-cueing hierarchy and homework exercises that utilized semantic, orthographic, and phonological cues. Despite a relatively rapid cognitive decline over the treatment period, LV demonstrated significantly improved naming on trained items, as well as generalization to untrained items and standardized tests such as the *Boston Naming Test* (*BNT*; Kaplan, Goodglass, & Weintraub, 1983) and *Western Aphasia Battery* (*WAB*; Kertesz, 1982). While no formal analyses were conducted and slight differences existed in treatment methods, the effect sizes (d statistics = 7.55 for LV and 7.22 for the other participant) for both participants in the study were similar, suggesting that “treatment administered via video conference may be a viable alternative for treating individuals who otherwise cannot be seen by a clinician,” (Henry et al., 2013b).

Although research investigating the use of teletherapy in PPA is scarce, research concerning teletherapy outcomes in stroke-related aphasia has established the feasibility and efficacy of the delivery method with a growing body of literature. Initial studies focused on the use of audio and video technology in the assessment of acquired communication disorders (Georgeadis, Brennan, Barker, & Baron, 2004; Hill, Theodoros, Russell, Ward, & Wootton, 2009; Theodoros, Hill, Russell, Ward, & Wootton, 2008; Vestal, Smith-Olinde, Hicks, Hutton, & Hart, 2006). Results of these studies demonstrated strong agreement between in person and remote assessment in evaluating individuals with stroke-related aphasia, as well as other acquired disorders such as traumatic brain injury and Alzheimer's disease.

In more recent studies, the focus has shifted to the use of audio and video technologies in the treatment of language disorders, including stroke-related aphasia. The few treatment studies available have focused on various linguistic deficits associated with aphasia, such as lexical retrieval (Agostini et al., 2014; Dechêne et al., 2011; Furnas & Edmonds, 2014; Fridler et al., 2012), sentence production (Goldberg, Haley, & Jacks, 2012), and apraxia (Lasker, Stierwalt, Spence, & Calvin-Root, 2010). The majority of studies have addressed lexical retrieval deficits. Dechêne et al. (2011) conducted a teletherapy study with three individuals with aphasia and anomia. The protocol targeted auditory and written production and comprehension of single words with various lexical tasks (e.g. repetition, oral reading, word to picture matching). Following treatment, all three participants showed modest gains in trained items compared to untrained items, and highly rated their satisfaction with the teletherapy medium. However, no control group was included to compare gains across conditions. Fridler et al. (2012) utilized a crossover design involving eight participants with chronic aphasia. In both the in person and remote phases of the study, sessions consisted of a probe of all trained items followed by language tasks, such as semantic elaboration, to improve naming. The participants' improvement, as measured by the aphasia quotient of the *WAB* (Kertesz, 1982), was found to be significantly greater following teletherapy regardless of treatment order. Similarly, Agostini et al. (2014) found comparable gains between both teletherapy and traditional therapy in a study that employed a crossover design with five participants with chronic aphasia. In both treatment phases, stimuli were presented on a computer screen to be named. If the item was not named, progressive phonemic cues were provided. Following treatment, significant effects were observed for time but no significant difference was found between teletherapy and in person therapy. Lastly, Furnas and Edmonds (2014) looked at the retrieval of verbs by delivering a verb network strengthening treatment to two individuals with chronic aphasia via teletherapy. Following eight weeks of treatment, both participants showed improvement in retrieving both trained and untrained words in sentence production, with generalization to untrained nouns and verbs. However, similar to Dechêne et al. (2011), no in person treatment group was included to analyze potential differences in treatment outcomes across conditions.

Fewer studies have looked at interventions addressing linguistic deficits such as syntax, discourse, or apraxia. Goldberg et al. (2012) implemented a treatment targeting sentence production by training individualized scripts in two participants with aphasia, with a combination of in-person and teletherapy sessions. Script training involved a progression through repetition tasks, choral reading, and independent production. Positive effects were noted for increased rate of speech, increased percent of target script words used, and decreased number of disfluencies. The authors concluded that the use of teletherapy did not influence the rate of learning negatively and that teletherapy is a viable method of delivering script training intervention when supported by occasional in-person sessions. Lasker, Stierwalt, Spence, and Calvin-Root (2010) also combined in-person and teletherapy sessions in a feasibility study to address severe apraxia in an individual with chronic apraxia and aphasia following stroke. The intervention utilized a motor learning guided approach with imitation, immediate and delayed repetition, reading aloud, and home practice with a speech generating device to increase production accuracy of target words and phrases. Following treatment, similar outcomes were observed between items treated via teletherapy and in person in terms of intelligibility, immediacy, and naturalness ratings, as well as an improvement from a rating of severe to moderate apraxia on the Apraxia Battery for Adults-2 (ABA-2; Dabul, 2000). Similar to Goldberg et al. (2012), Lasker et al. (2010) concluded that their initial results suggest that teletherapy can be as effective as in-person therapy. Overall, the current literature on stroke-related aphasia and teletherapy indicates that gains in teletherapy are at least comparable if not greater than those observed following traditional face-to-face treatment.

1.5 Current Study

Efficacy of teletherapy in stroke-related aphasia has been established by a small but growing body of research that compares treatment outcomes in teletherapy to outcomes in traditional face-to-face therapy. However, this research has not been extended to PPA, as only one study was identified that addresses teletherapy outcomes in individuals with PPA. The aim of this study was to compare treatment outcomes in teletherapy and traditional therapy in a group of individuals with PPA to determine whether comparable gains are achieved when intervention is delivered via teletherapy. Further, we sought to examine whether differences in generalization or maintenance of gains existed between teletherapy and traditional therapy. Existing data from the Aphasia Research and Treatment Laboratory at the University of Texas at Austin and the University of California San Francisco Memory and Aging Center were used in the data analysis. The groups of participants included all three PPA variants and consisted of two therapy protocols; a script training protocol for nfvPPA participants and a lexical retrieval protocol for svPPA and lvPPA participants. We hypothesized that teletherapy outcomes would be comparable to traditional forms of treatment. If our hypothesis is correct, this holds significant implications for treatment outcomes and increased access to speech language pathologists that are qualified for and familiar with treating these relatively rare variants of neurodegenerative disease.

2. Methods

2.1 Participants and Assessment Measures

Participants included in the data analysis were individuals referred for a comprehensive neurological, neuropsychological, and speech-language evaluation at either the Aphasia Research and Treatment Laboratory at the University of Texas at Austin or the University of California San Francisco Memory and Aging Center, and then subsequently enrolled in a speech-language treatment study. To qualify for entry into a treatment study, participants with language impairment had to meet current diagnostic criteria for PPA (Gorno-Tempini et al., 2011). A PPA diagnosis required insidious onset and gradual deterioration of speech and language functions, with speech and language deficits comprising the most prominent deficits throughout the early stages of disease. Patients were diagnosed with svPPA, lvPPA, or nfvPPA based on current guidelines (Gorno-Tempini et al., 2011). Diagnosis according to variant was made by consensus following a review of the patient's medical history, a multidisciplinary evaluation encompassing language and neuropsychological testing (see Henry et al., 2016 for overview of assessment procedures), and neurological exam.

A total of 25 individuals with PPA were included (8 with svPPA, 8 with lvPPA, and 9 with nfvPPA). At the time of the data analysis, all participants had completed their program of treatment and a post-treatment assessment. A majority of the participants (22 individuals; 7 with svPPA, 6 with lvPPA, and 9 with nfvPPA) had also completed three month follow-up assessments. In addition, a smaller subset of participants had completed six month follow-up assessments (17 individuals; 4 svPPA, 5 lvPPA, 8 nfvPPA). Participants were assigned to teletherapy versus traditional face-to-face therapy according to patient need (i.e. travel concerns, mobility issues, or residence in a different city or state). Of the 25 participants, 16 received traditional therapy and 9 received teletherapy.

See Tables 1 and 2 for demographic characteristics and relevant speech and language measures from initial, post-treatment, and follow-up assessments.

Table 1. Demographic Characteristics (with standard deviations) for Participants According to Treatment Protocol and Type

Demographic	LRT Face-to-Face *(n = 11)*	LRT Teletherapy *(n = 5)*	VISTA Face-to-Face *(n = 5)*	VISTA Teletherapy *(n = 4)*
PPA variant	5 svPPA:6 lvPPA	3 svPPA:2 lvPPA	5 nfvPPA	4 nfvPPA
Age	68.1 (6.8)	56.8 (5.0)	67.60 (3.8)	68.75 (8.0)
Sex	4M:7F	3M:2F	2M:3F	1M:3F
Education (years)	17.8 (2.8)	17.0 (3.3)	15.60 (2.6)	16.25 (1.3)
Handedness	8R;1L;2Amb	4R;1L	5R	4R
MMSE (30)	24.0 (4.4)	23.6 (4.6)	26.00 (2.3)	28.50 (0.6)

LRT = Lexical Cascade Retrieval Treatment, VISTA = Video-Implemented Script Training in Aphasia (see section 2.2 Treatments for discussion)

M = male, F = female, R = right, L = left, Amb = ambidextrous.

MMSE = Mini Mental State Examination

Table 2. Group Mean Performance (with standard deviations) on Relevant Speech and Language Measures According to Treatment Protocol and Type

	LRT Face-to-Face (*n = 11*)	LRT Teletherapy (*n = 5*)	VISTA Face-to-Face (*n = 5*)	VISTA Teletherapy (*n = 4*)
Pre-Tx BNT (%)[b]	40.9 (24.4)	34.5 (26.2)	--	--
Post-Tx BNT (%)[b]	44.5 (22.6)	38.5 (28.4)	--	--
3 month BNT (%)[b]	43.9 (22.3) (*n = 9*)[a]	37.5 (30.7) (*n = 4*)[a]	--	--
6 month BNT (%)[b]	44.0 (18.0) (*n = 5*)[a]	40.0 (29.1) (*n = 3*)[a]	--	--
Pre-Tx NAT (%)[b]	--	--	58.3 (25.0) (*n = 4*)[a]	70.8 (22.2)
Post-Tx NAT (%)[b]	--	--	71.3 (20.9)	85.0 (13.7)
3 month NAT (%)[b]	--	--	64.7 (23.4)	75.0 (34.0)
6 month NAT (%)[b]	--	--	44.2 (36.3) (*n = 4*)[a]	66.7 (38.1) (*n = 3*)[a]
Pre-Tx WAB AQ (100)	80.1 (10.7)	81.7 (8.5)	82.9 (5.7)	88.0 (5.9)
Post-Tx WAB AQ (100)	80.4 (10.7)	82.9 (6.3)	83.8 (5.7)	89.4 (5.8)
3 month WAB AQ (100)	81.0 (10.0) (*n = 9*)[a]	79.0 (9.5) (*n = 4*)[a]	78.5 (8.2)	87.0 (7.3)
6 month WAB AQ (100)	76.8 (11.0) (*n = 6*)[a]	75.0 (15.2) (*n = 3*)[a]	76.7 (7.1)	85.8 (12.1) (*n = 3*)[a]

[a] Differences in *n* as not all participants had completed follow up assessments or due to missing data

[b] Generalization measures were selected according to deficits targeted by the treatment protocol. The *BNT* was used to examine generalization in LRT while the *NAT* examined generalization in VISTA.

LRT = Lexical Cascade Retrieval Treatment, VISTA = Video-Implemented Script Training in Aphasia (see section 2.2 Treatments for discussion)

BNT = Boston Naming Test; *NAT* = Northwestern Anagram Test; *WAB AQ* = Western Aphasia Battery Aphasia Quotient

2.2 Treatment Approaches

The treatment approach implemented for each individual varied by PPA variant in order to address core linguistic deficits characteristic of each subtype of PPA. A total of three treatment approaches were used to treat participants: 1) the original version of Lexical Retrieval Cascade Treatment (LRT-1; 5 individuals with svPPA and 5 individuals with lvPPA), 2) a modified version of Lexical Retrieval Cascade Treatment (LRT-2; 3 individuals with svPPA and 3 individuals with lvPPA), and 3) Video-Implemented Script Training in Aphasia (VISTA; 9 individuals with nfvPPA). All treatment approaches, whether conducted face-to-face or via video conferencing software, utilized the same protocols and materials. Each treatment approach is discussed in greater detail below. See Table 3 for number of participants in each protocol and number of participants within each approach receiving face-to-face therapy versus teletherapy.

Table 3. Number of Participants within Each Treatment Group Receiving Face-To-Face Therapy vs. Teletherapy

Treatment Approach	LRT-1	LRT-2	VISTA
Total number of participants	5 svPPA:5 lvPPA	3 svPPA:3 lvPPA	9 nfvPPA
Individuals receiving face-to-face therapy	3 svPPA:4 lvPPA	2 svPPA:2 lvPPA	5 nfvPPA
Individuals receiving teletherapy	2 svPPA:1 lvPPA	1 svPPA:1 lvPPA	4 nfvPPA

2.2.1 Lexical Retrieval Cascade Treatment (LRT)

The treatment approach implemented for ten of the sixteen participants with svPPA or lvPPA is referred to as Lexical Retrieval Cascade Treatment (LRT-1), an approach developed at the University of Arizona to facilitate word retrieval (Henry et al., 2013b). With this approach, participants are trained to use lexical retrieval strategies by capitalizing on residual semantic, orthographic, and phonological knowledge. The treatment utilizes a cueing hierarchy that guides the participant through a series of tasks to strengthen and activate central components of language processing, while also training self-cueing techniques. Table 4 summarizes the cueing hierarchy used in treatment.

Table 4. Lexical Retrieval Cueing Hierarchy (from Henry et al., 2013b)

(Picture is presented) 1. Semantic self-cue	Clinician prompts semantic description with "Tell me about this object." • Additional prompts are given as needed, such as "What does it look like? Where can you find it? What do you use it for?" • For participants with degraded semantic knowledge, prompts are given for description of personal experience or memories with the object, such as "What memories do you have about this?"
2. Orthographic self-cue	Clinician requests written form of the word with "Can you write the word?" • If unable to write the word, clinician encourages participant to think of the first letter and/or sound of the word or any other characteristics about the word (i.e. "Is it a long or short word?") • If the participant is unable to come up with any orthography, the clinician provides the first letter and prompts, "Does this help you say the word?"
3. Phonemic self-cue	Clinician prompts participant to say the sound that the letter makes with "Make the sound of this letter. Now try to say the word."

Table 4 (continued)

4. Oral reading	If the item is not yet named, the clinician writes out the remainder of the word and the participant reads it out loud.
5. Spoken and Written Repetition	Clinician asks participant to write and say the word three times. • Written model remains in sight and incorrect written attempts are crossed out.
6. Semantic Plausibility Judgments	Clinician asks five yes/no questions regarding semantic features of the item, such as "Is this something you find in the kitchen?" • Clinician removes written production attempts and places picture back in front of participant • Clinician corrects any inaccurate responses.
7. Recall	Clinician asks participant to recall two features by prompting "Tell me what are the two most important features of this object." Clinician asks the participant to write and say the word one time.

In addition to treatment sessions, participants completed daily homework based on Copy and Recall Treatment (CART; Beeson & Egnor, 2006). This involved repeated copy and spoken production of target words (ten times), followed by recall of spoken and written word forms from memory.

2.2.1.1 Stimulus Selection in LRT

Twenty imageable nouns were targeted for training and five nouns were selected to remain untrained. Stimuli were selected from a larger set of items according to performance on baseline probes, as well as utility of the target word for the participant. Only items that were not named correctly on at least two of three probing opportunities were included for treatment. Once 25 items were established, items were divided into four trained and one untrained sets of five, which were matched for relevant linguistic characteristics such as familiarity, frequency, number of letters, number of syllables, and imageability. Item characteristics were obtained from the Corpus of Contemporary American English (Davies, 2008-) and MRC Psycholinguistic Database (Coltheart, 1981). In creating stimulus materials, participants with svPPA provided personal photos of items to aid generalization to the home environment due to item- and context-specific treatment gains demonstrated in these individuals (Jokel et al., 2014). Participants with lvPPA used generic photos of items from a large database.

2.2.1.2 Training Criteria and Data Collection in LRT

The twenty items were trained in four sets of five using a multiple baseline design in which one set was treated per session. Treatment sessions occurred once a week for one hour and each set was trained for a minimum of one and a maximum of two sessions with the clinician, until 80% items were correctly named. In accordance with mastery criteria, participants underwent a minimum of four weeks of treatment (4 sessions, total of 4 hours) up to a maximum of eight weeks of treatment (8 sessions, total of 8 hours). Performance on all trained and untrained sets was probed at the beginning of each treatment session so that all item sets were probed once per week. During probing, no cuing was provided by the clinician, and all items named spontaneously or with participant-initiated cues were scored as correct. Generalization was examined via performance on untrained items, as well as the *Boston Naming Test* (*BNT*; Kaplan et al., 1983) and *WAB Aphasia Quotient* (*WAB AQ*; Kertesz, 1982).

2.2.2 Modified Version of Lexical Retrieval Cascade Treatment

Six of the sixteen participants with svPPA or lvPPA underwent a modified version of LRT (LRT-2). This version utilized the same protocol employed in the previous version (see Table 4 for summary) but differed only in treatment dosage and number of stimuli. Treatment sessions occurred twice per week for one hour, with each set trained for a minimum of one session and a maximum of two session until 100% of items in the set were correctly named. Thus, individuals participated in treatment for a minimum of 4 weeks (8 sessions, total of 8 hours) and a maximum of 8 weeks (16 sessions, total of 16 hours). Treatment also targeted a larger set of items. Forty nouns were trained in eight sets of five, while an additional ten nouns remained untrained. Due to the larger number of items, performance on the set being currently treated and half of all other trained and untrained sets was probed at the beginning of each treatment session so that all item sets were probed once per week. All other variables (stimulus development, treatment cueing hierarchy and protocol, data collection, etc.) remained the same between the two versions.

2.2.3 Video-Implemented Script Training in Aphasia (VISTA)

Participants with nfvPPA underwent Video-Implemented Script Training in Aphasia (VISTA), a novel treatment approach designed to facilitate grammaticality, intelligibility, and fluency of connected speech. This treatment employs script training, an approach that involves repeated practice of phrases or sentences in either a monologue or dialogue (Cherney, Halper, Holland & Cole, 2008; Youmans, Holland, Munoz & Bourgeois, 2005). VISTA is a largely homework-based protocol that is implemented via "speech entrainment," a relatively new technique in aphasia treatment that utilizes repeated practice with an audiovisual model of a healthy speaker, which participants attempt to mimic in real time (Fridriksson et al., 2012). This daily practice is complemented by sessions with the clinician that target articulatory and grammatical aspects of script production. In addition to targeting speech production skills, sessions also target memorization and conversational usage of scripted materials via a treatment hierarchy that moves from structured to more functional tasks. Table 5 summarizes the treatment hierarchy.

Table 5. VISTA Clinician-guided Treatment Hierarchy

Treatment Steps	
Structured Treatment	1. Recall/Recognize • Clinician asks participant to choose the correct trained script sentence from four foil sentences
	2. Put script sentences in order • After selecting all the correct sentences from foils, the clinician asks participant to put them in order of the script.
↓	3.Read script aloud
	4. Produce script sentences in response to questions (in scripted order) • Clinician prompts each sentence with a question (e.g. "Where were you born?") • If necessary, clinician cues for complete sentence.
	5. Produce entire script from memory • Clinician prompts for complete script with "Tell me about…"
Functional Application	6. Respond to questions with scripted sentences (not in scripted order) • Clinician asks questions on the scripted topic in a conversation and elicits scripted sentences out of order.

*Notes: Feedback regarding articulation and grammar occur during steps 3-6, with targeted practice as needed. During the second treatment session for a given script, a novel communication partner has an unscripted conversation with the participant to promote conversational usage of scripted material.

In addition to treatment sessions with the clinician, homework consisted of unison speech production practice with the video model for a minimum of 30 minutes per day.

2.2.3.1 Stimulus Selection in VISTA

Treatment stimuli consisted of four trained scripts and two untrained scripts ranging from four to six sentences in length. Scripts were tailored to each participant and were developed via a collaborative process between the participant, caregiver, and clinician. Participants chose script topics and generated sentences, which the clinician then edited to ensure the script was appropriate for treatment (e.g. limited use of multisyllabic words due to articulatory difficulties). Following script development, video stimuli were created with a healthy speaker of the same sex as the participant. The healthy speaker read the participant's script at a tailored rate while exaggerating articulatory gestures to provide salient cues for production. Speech rate was determined by deriving the participant's words produced per minute during a picture description task and a reading passage. The faster rate between the two tasks was selected to ensure a natural, attainable rate that was still challenging for the participant. Scripts were balanced for number of words, number of sentences, complexity, readability, and Flesch-Kincaid Grade Level (Kincaid, Fishburne, Rogers, & Chissom, 1975).

2.2.3.2 Training Criteria and Data Collection in VISTA

The four scripts were trained using a multiple baseline design, with treatment sessions occurring twice per week for an hour. Each script was trained for a minimum of two and a maximum of three sessions with the clinician until 90% of scripted words were produced correctly and intelligibly. Treatment lasted a minimum of 4 weeks (8 sessions, total of 8 hours) up to a maximum of 6 weeks (12 sessions, total of 12 hours). A correct production of a word required that it was a lexical unit from the script; intelligibility was defined as whether a naïve listener could understand the target word within the context of the script. Performance on the script being currently treated and half of all other trained and untrained scripts was probed at the beginning of each treatment session so that all scripts were probed once per week. Generalization was examined by percent correct intelligible words on untrained sets, as well the *Northwestern Anagram Test* (*NAT*: Weintraub et al., 2009), a nonverbal test of syntactic production, and the *WAB AQ* (Kertesz, 1982).

2.3 Primary Outcome Measures

The primary outcome measure was the pre- to post-treatment change scores in the dependent variable for each treatment approach. In the LRT-1 and LRT-2 cases, this was defined as the difference between average percent correct from the last two pre-treatment probes relative to average percent correct from the first two post-treatment probes (calculated for both trained and untrained sets). With the VISTA cases, this was defined as the difference between average percent correct intelligible words from the last two pre-treatment probes relative to the average percent correct intelligible words from the first two post-treatment probes (calculated for both trained and untrained scripts). Maintenance change scores (the difference between performance at three months as well as six months relative to post-treatment) for trained and untrained stimuli were also calculated for a subset of participants (22/25 individuals for three month change scores, 17/25 individuals for six month change scores) in order to examine maintenance of treatment gains.

Generalization of treatment effects to standardized tests was also examined via change scores as a secondary outcome measure. Both pre- to post-treatment change scores and maintenance change scores were calculated for relevant standardized tests according to treatment approach. When a discrepancy occurred in the version administered between the two time points (e.g. administration of 60 item *BNT* at post-treatment and 30 item *BNT* at 3 months post-treatment), equivalent scores were calculated by reviewing the scoring protocol and calculating the score on the same set of items, to ensure valid comparison across time points. Generalization in LRT-1 and LRT-2 cases was examined via the *BNT* and the *WAB AQ*, whereas in the VISTA cases, this was examined via the *NAT* and the *WAB AQ*.

2.4 Statistical Analysis

Statistical analysis was performed using IBM SPSS Statistics (version 23.0, Chicago, IL, USA). A significance level of 0.05 was used in all analyses. Given that normal distribution and homogeneity of variances could not be assumed due to small sample size, variables were analyzed and compared using nonparametric statistical procedures. Furthermore, to achieve adequate statistical power to analyze LRT outcomes, we combined LRT-1 and LRT-2 data. In order to establish whether the two groups were comparable, demographic and clinical variables were examined for any differences, so that interpretation of results could take into account any variables that could introduce additional variation in treatment outcomes within the group. Group differences between LRT-1 and LRT-2 were examined using a nonparametric Mann-Whitney U test for age, education, *MMSE*, and pre-post change scores on trained and untrained treatment items.

Data were first analyzed to determine if the treatment approach used (i.e. LRT, VISTA) produced a significant treatment effect over time, regardless of treatment type (face-to-face vs. teletherapy). Wilcoxon signed-ranks tests were conducted to compare pretreatment performance with post-treatment performance on trained and untrained items, as well as standardized tests. Wilcoxon test statistics for pre-post treatment analysis are reported using a 1-tailed test, as we predicted improvement following treatment. Maintenance effects were also examined with Wilcoxon signed-ranks tests, which compared performance at post-treatment with follow-up performance at three and six months on trained and untrained items and standardized tests. Wilcoxon test statistics for post-treatment to follow-up assessments are reported using a 2-tailed test, as improvement during this period could not be assumed. These analyses are reported separately for both LRT and VISTA protocols, as the treatments target different core linguistic skills.

Subsequently, data were analyzed to determine whether the magnitude of change following treatment differed according to treatment type (i.e. face-to-face vs. teletherapy) within each approach. To examine whether differences existed, the Mann-Whitney U test was used to compare pre-post treatment change scores (% change) in face-to-face participants versus teletherapy participants. Likewise, differences in maintenance for treatment type were examined via Mann-Whitney U tests that compared post-treatment to three month follow-up change scores and post-treatment to six month follow-up change scores between the treatment types. All analyses were conducted using change scores for both trained and untrained items, as well as change scores for standardized tests. All Mann-Whitney U test statistics were reported using an exact sampling distribution for *U* (Dineen & Blakesley, 1973).

3. Results

3.1 Statistical Analysis of LRT Data

3.1.1 Establishment of Single LRT Participant Group

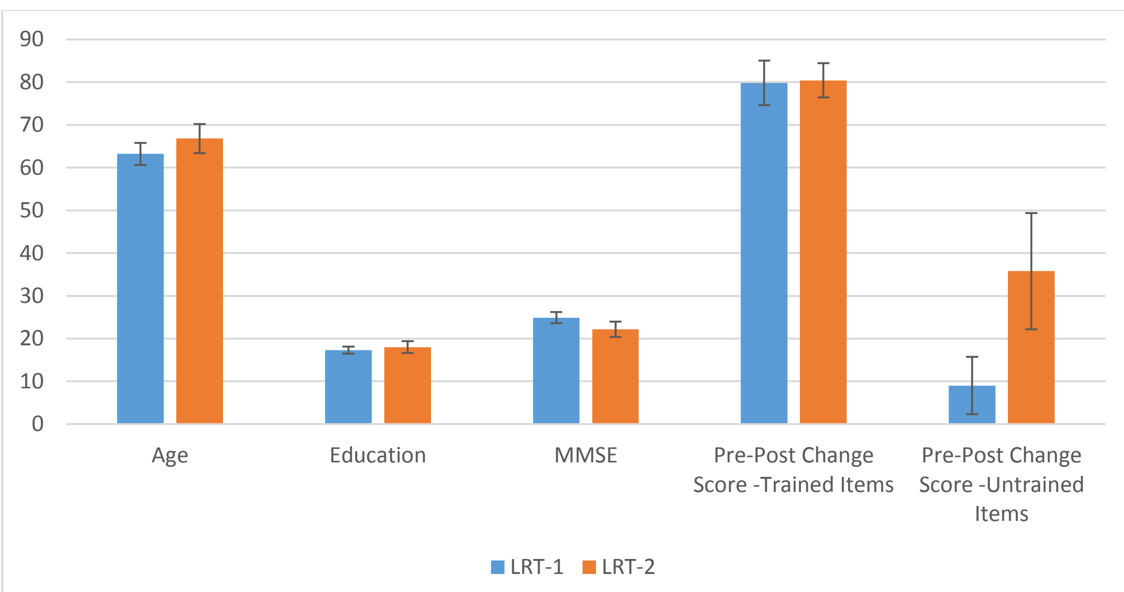
Demographic variables and pre-post change scores for trained and untrained items in participants receiving LRT-1 and participants receiving LRT-2 are presented in Table 6 and Figure 1. A Mann-Whitney U test was conducted to determine if differences existed between the two LRT conditions in age, education, *MMSE*, or pre-post change scores on trained and untrained items. The two groups were not significantly different for age ($U = 20.50$, $z = -1.03$, $p = .313$), education ($U = 23.50$, $z = -.716$, $p = .492$), *MMSE* ($U = 16.50$, $z = -1.481$, $p = .147$), pre-post change score for trained items ($U = 26.50$, $z = -.380$, $p = .713$), or pre-post change score for untrained items ($U = 14.00$, $z = -1.748$, $p = .093$) using an exact sampling distribution for U (Dineen & Blakesley, 1973).

Given that no significant differences in demographic or clinical variables were found between the two groups, data from both LRT groups were combined into one dataset (LRT-All) for all subsequent analyses. This resulted in a group of 16 participants, with 11 receiving traditional face-to-face therapy and 5 receiving teletherapy.

Table 6. Mean Demographic Characteristics and Pre-Post Change Scores for Trained and Untrained Items (with standard deviations) in LRT-1 and LRT-2 Participants.

	LRT-1 (*n = 10*)	LRT-2 (*n = 6*)
Age	63.2 (8.2)	66.8 (8.4)
Education	17.3 (2.6)	18.0 (3.5)
MMSE	24.9 (4.2)	22.2 (4.4)
Pre-Post Change Score (%) Trained Items	79.8 (16.4)	80.4 (9.9)
Pre-Post Change Score (%) Untrained Items	9.0 (21.3)	35.8 (33.4)

Figure 1. Mean Demographic Characteristics and Pre-Post Change Scores for Trained and Untrained Items in LRT-1 and LRT-2 Participants. Error bars represent standard errors.



* = $p < 0.05$

3.1.2 Treatment Effect and Generalization Following LRT

Group mean performance on all pre- and post-treatment measures is presented in Table 7 and Figure 2. All test statistics reported for measuring treatment effect and generalization were obtained from a Wilcoxon signed ranks test using a 1-tailed test. All 16 participants saw improvement in naming trained items following LRT, with nine participants also seeing improvement in naming untrained items following treatment. This change constituted a significant treatment effect for trained items (median change in percent correct of +83.13%; $z = -3.517$, $p = .0005$) as well as a significant generalization effect to untrained items (median change in percent correct of +12.50%, $z = -2.385$, $p = .009$).

In examining generalization to standardized tests following LRT, 11 of the participants also saw improvement on the *BNT*, while four saw a decline in naming on this measure from pre- to post-treatment. At the group level, analysis revealed that generalization to performance on the *BNT* was significant (median change of +5.00%; $z = -2.046$, $p = .020$). Results were mixed on the *WAB AQ*, with eight participants showing improvement and eight showing deterioration in their score. No significant change in *WAB AQ* was observed (median change in *WAB AQ* of +.15; $z = -.207$, $p = .418$).

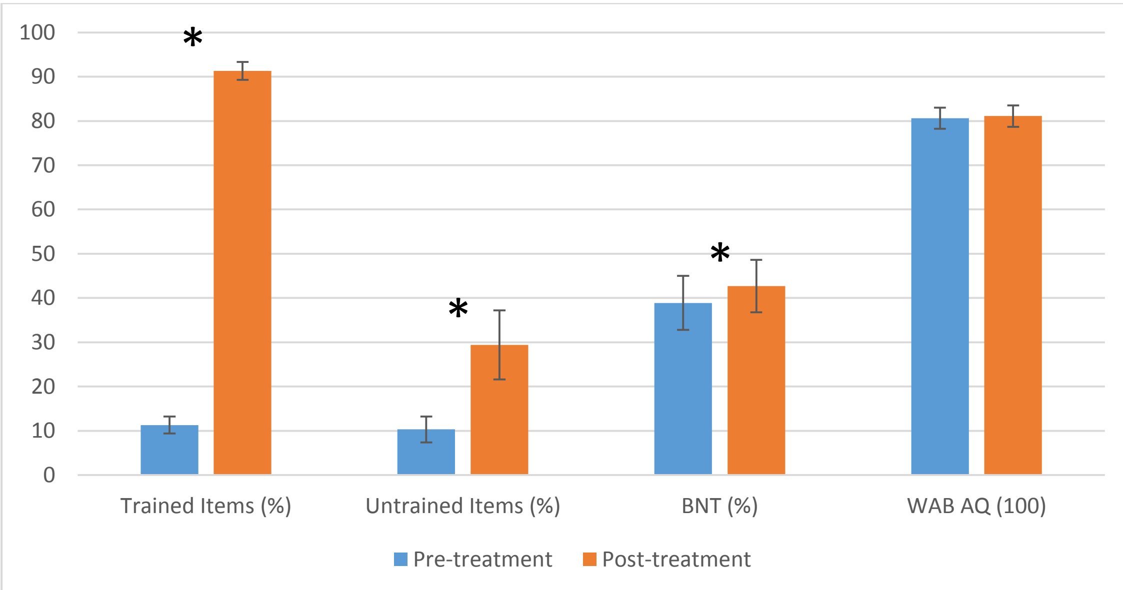
Table 7. Mean Performance on All Dependent Variables (with standard deviations) for All LRT Participants

	Pre-treatment (*n = 16*)	Post-treatment (*n = 16*)	Post-treatment Equivalent [a] (*n = 14*)	3 Month Follow-up (*n = 13*)	6 Month Follow-up (*n = 9*)
Trained Items (%)	11.3 (7.6)	91.3 (8.0)	--	84.6 (14.4)	88.3 (12.0)
Untrained Items (%)	10.3 (11.6)	29.4 (31.1)	--	32.3 (30.3)	15.6 (26.0)
BNT (%)	38.9 (24.3)	42.7 (23.7)	46.9 (20.8)	41.9 (24.0)	42.5 (24.0) (*n = 8*)[b]
WAB AQ (100)	80.6 (9.8)	81.1 (9.4)	--	80.4 (9.5)	76.2 (11.6)

[a] The full 60 item *BNT* was administered at pre- and post-treatment but a shortened version (15 or 30-item) was administered at 3 and 6 month follow-up. For each participant with follow-up data, a Post-equivalent *BNT* was calculated by reviewing the scoring protocol and calculating the score on the same set of items, to ensure for valid comparison across time points.
[b] Due to missing data

BNT = Boston Naming Test; *WAB AQ* = Western Aphasia Battery Aphasia Quotient

Figure 2. LRT Group Mean Performance on All Pre- and Post-treatment Measures. Error bars represent standard errors.



* = p < 0.05

3.1.3 Maintenance of Gains Following LRT

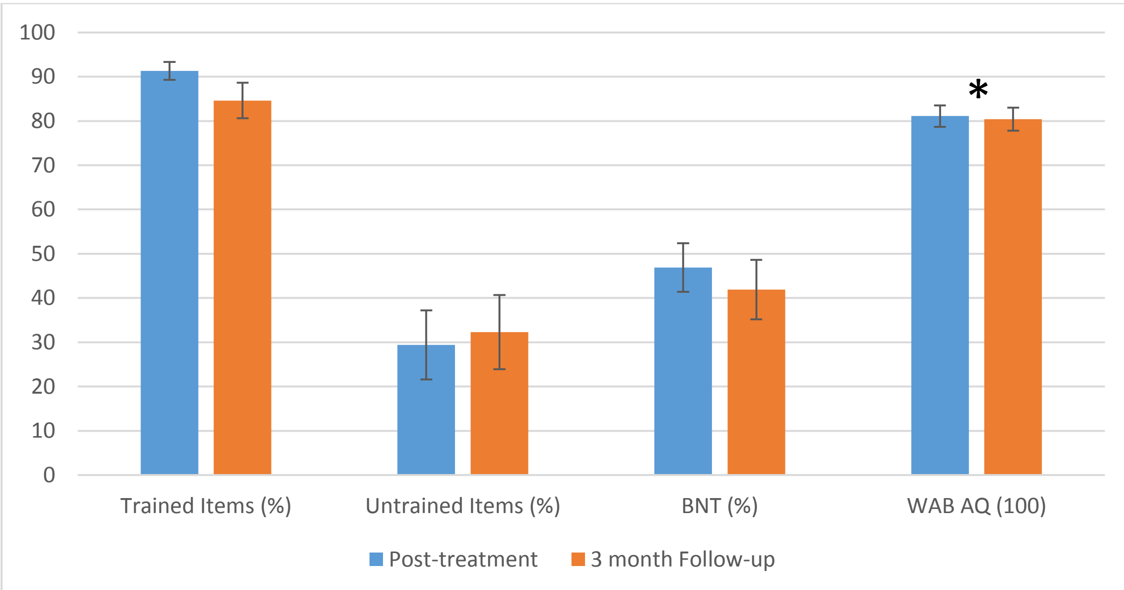
Group mean performances on all post-treatment, three month follow-up, and six month follow-up measures are presented in Table 7 and Figures 3 and 4. All test statistics reported for measuring maintenance of gains were obtained from a Wilcoxon signed ranks test using a 2-tailed test. Of the original 16 participants, 13 participants were included in the analysis of maintenance of gains from post-treatment to three month follow-up testing. During this time nine of the 13 participants experienced a decline in naming trained items. However, this decline was not significant (median change in percent correct of -2.5%; z = -1.921, p = .055). Similarly, gains were also were maintained at three months follow-up on untrained items, with no significant decline found (median change in percent correct of 0.0%; $z = -.085$, $p = .933$).

Maintenance of gains on standardized tests at three months was also examined for both the *BNT* and *WAB AQ*. At three months follow-up, the majority of participants showed either a slight decline (6/13) or no change (3/13) in their *BNT* score, changes that were not found to be significant (median change of 0.0%; $z = -.919$, $p = .358$). However, significant decline was found on the *WAB AQ* (median change of -2.0 points; $z = -2.273$, $p = .023$), with ten of the 13 participants scoring lower at three months follow-up than at post-treatment.

Nine of the original 16 participants were also included in the analysis of maintenance from post-treatment to six month follow-up testing. While approximately half of the participants showed decline in naming trained and untrained items (5/9 and 4/9 participants respectively), neither trained (median change of -5.00%; $z = -1.272$, $p = .203$) or untrained items (median change of 0.0%; $z = -.431$, $p = .666$) showed significant decline from post-treatment to six months follow-up.

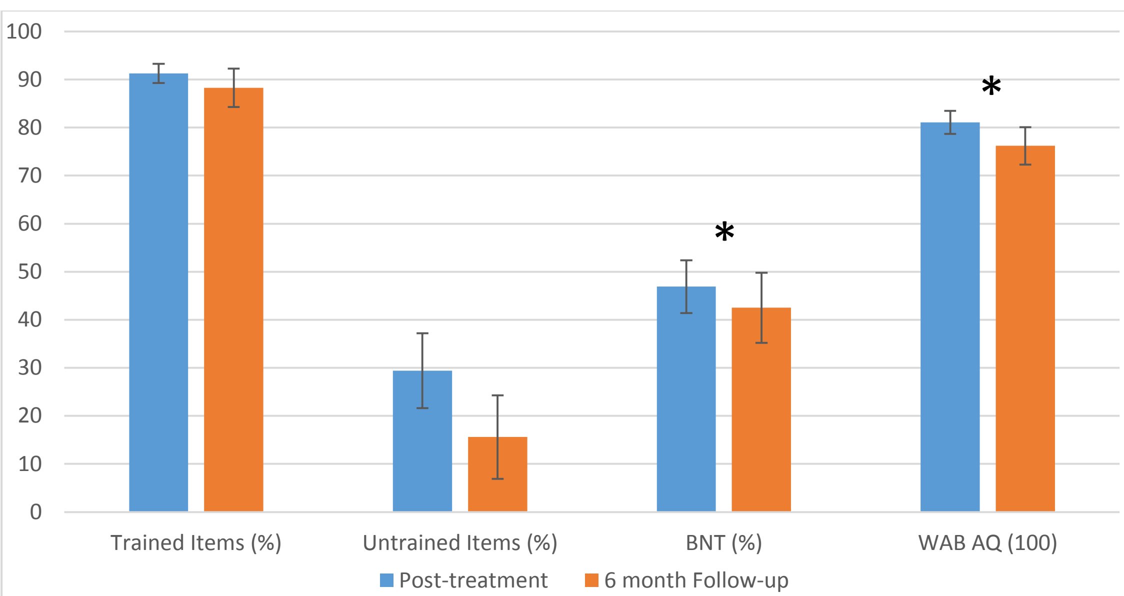
From post-treatment to six months follow-up, almost all participants showed decline on the *BNT* and the *WAB AQ* (7/8 and 8/9 participants respectively). Overall, this deterioration in scores was significant for both the *BNT* (median change of -6.67%; $z = -2.388$, $p = .017$) and the *WAB AQ* (median change of -3.5 points; $z = -2.521$, $p = .012$).

Figure 3. LRT Group Mean Performance on Post-treatment and 3 Month Follow-up Measures. Error bars represent standard errors.



* = $p < 0.05$

Figure 4. LRT Group Mean Performance on Post-treatment and 6 Month Follow-up Measures. Error bars represent standard errors.



* = $p < 0.05$

3.1.4 Treatment Effect in Traditional Face-to-Face Therapy versus Teletherapy in LRT Participants

After examining all LRT cases together for treatment effects and maintenance of gains over time, data were analyzed for between-group differences in magnitude of change observed in LRT face-to-face cases versus LRT teletherapy cases. Mean percent change scores by treatment type on all pre- and post-treatment measures is presented in Table 8 and Figure 5. All test statistics reported for measuring between group differences were obtained from a Mann-Whitney U test using an exact sampling distribution for *U* (Dineen & Blakesley, 1973). While the two groups were not significantly different in education ($U = 25.0$, z = -.288, p = .827) or pre-treatment *MMSE* score ($U = 26.0$, $z = -.172$, $p = .913$), the two groups did differ significantly in age ($U = 8.5$, $z = -2.159$, $p = .027$), with participants in the LRT teletherapy group being younger (mean age of 56.8 years) compared to the LRT face-to-face group (mean age of 68.1 years).

Immediately following treatment, pre-post change scores for trained items for LRT face-to-face (mean rank = 9.41) and LRT teletherapy (mean rank = 6.50) were not significantly different ($U = 17.5$, $z = -1.134$, $p = .267$). Similarly, pre-post change scores for untrained items for LRT face-to-face (mean rank = 9.86) and LRT teletherapy (mean rank = 5.50) were not significantly different ($U = 12.5$, $z = -1.712$, $p = .090$). Generalization to both the *BNT* and the *WAB AQ* was also comparable between LRT face-to-face (mean rank = 8.32, 9.18 respectively) and LRT teletherapy (mean rank = 8.90, 7.00 respectively), as neither the pre-post *BNT* change score ($U = 25.5$, $z = -.227$, $p = .827$) or the pre-post *WAB AQ* change score ($U = 20.0$, $z = -.850$, $p = .441$) were significantly different between the two groups.

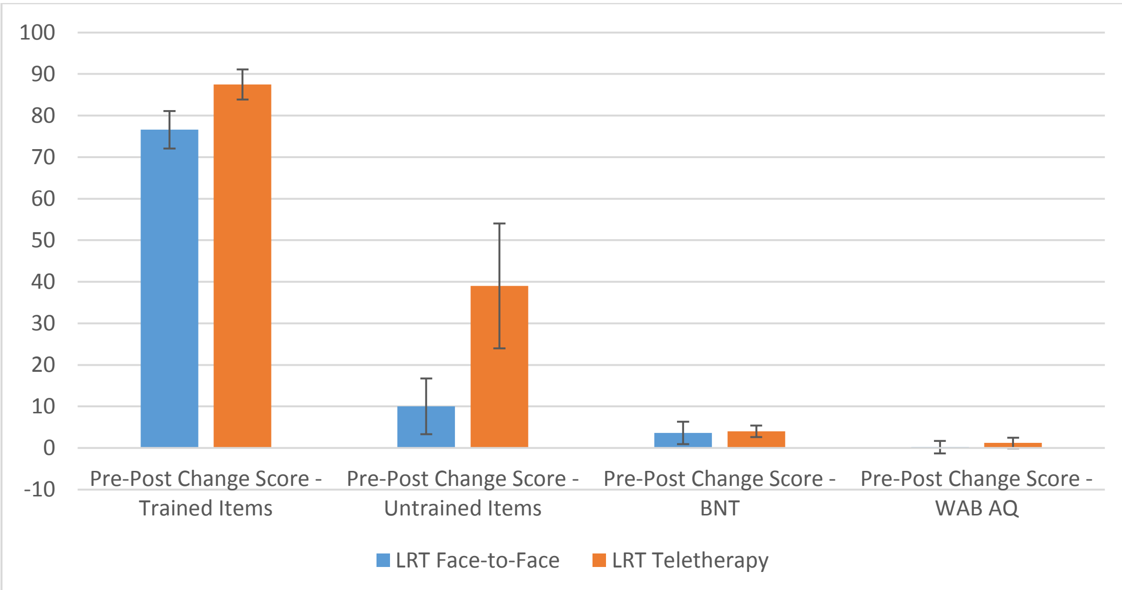
Table 8. Mean Change Scores (with standard deviations) for LRT Face-to-Face and LRT Teletherapy.

	Pre-Post Change Score (%)		**Post-3 Month Change Score (%)**		**Post-6 Month Change Score (%)**	
	LRT Face-to-Face (*n* = *11*)	LRT Teletherapy (*n* = *5*)	LRT Face-to-Face (*n* = *9*)	LRT Teletherapy (*n* = *4*)	LRT Face-to-Face (*n* = *6*)	LRT Teletherapy (*n* = *3*)
Trained Items	76.6 (15.0)	87.5 (8.1)	-9.4 (14.6)	-.94 (1.2)	-2.1 (12.0)	-12.5 (13.9)
Untrained Items	10.0 (22.2)	39.0 (33.6)	-4.4 (22.6)	17.5 (21.0)	-8.3 (24.0)	13.3 (57.7)
BNT	3.64 (8.8)	4.0 (3.0)	-6.1 (12.3)	.84 (16.4)	-10.7 (6.0) (*n* = 5)[a]	-4.4 (3.8)
WAB AQ	.24 (5.0)	1.2 (2.9)	-1.9 (3.0)	-3.1 (3.1)	-4.6 (3.8)	-7.3 (6.8)

[a] Due to missing data

BNT = Boston Naming Test; *WAB AQ* = Western Aphasia Battery Aphasia Quotient

Figure 5. LRT Face-to-Face Pre-Post Change Scores vs. LRT Teletherapy Pre-Post Change Scores. Error bars represent standard errors.



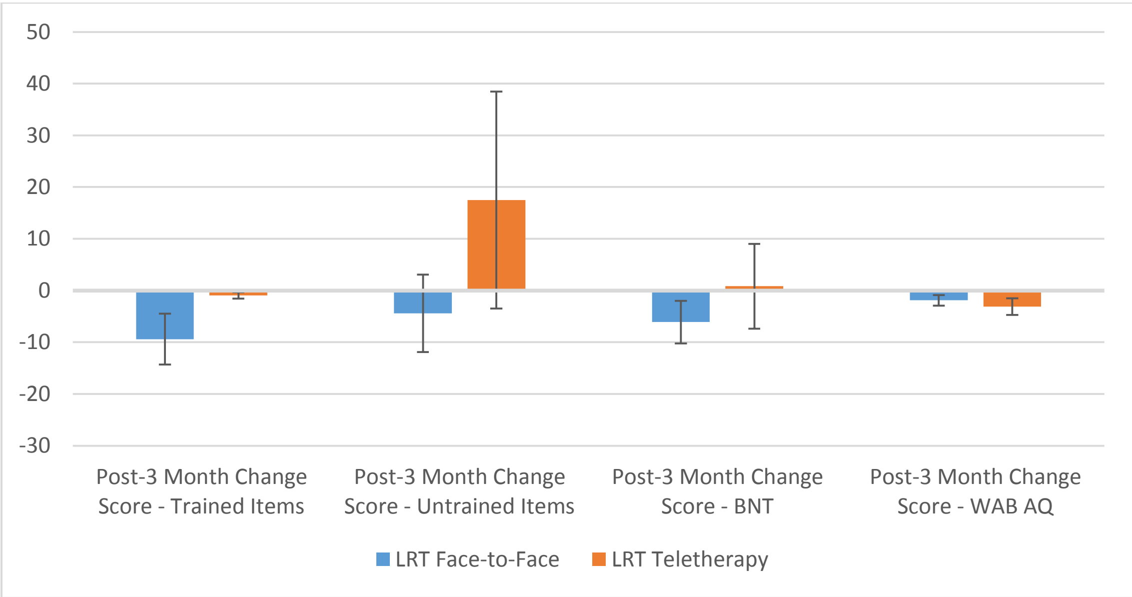
* = p < 0.05

3.1.5 Maintenance Following Traditional Face-to-Face Therapy versus Teletherapy in LRT Participants

Maintenance change scores from post-treatment to three months follow-up were also compared between the two treatment types. See Table 8 and Figure 6 for mean change scores from post-treatment to three month follow-up by treatment type. Post-treatment to three month follow-up change scores for trained items for LRT face-to-face (mean rank = 8.00) and LRT teletherapy (mean rank = 4.75) were not significantly different (U = 9.0, z = -1.398, p = .199). Similarly, post-treatment to three month follow-up change scores for untrained items for LRT face-to-face (mean rank = 7.44) and LRT teletherapy (mean rank = 6.00) were not significantly different (U = 14.0, z = -.649, p = .604). Maintenance on both the *BNT* and the *WAB AQ* was also comparable between LRT face-to-face (mean rank = 7.67, 6.44 respectively) and LRT teletherapy (mean rank = 5.50, 8.25 respectively), as neither the post-treatment to three month follow-up change score for the *BNT* (U = 12.0, z = -.932, p = .414) or the *WAB AQ* (U = 13.0, z = -.772, p = .503) were significantly different between the two groups.

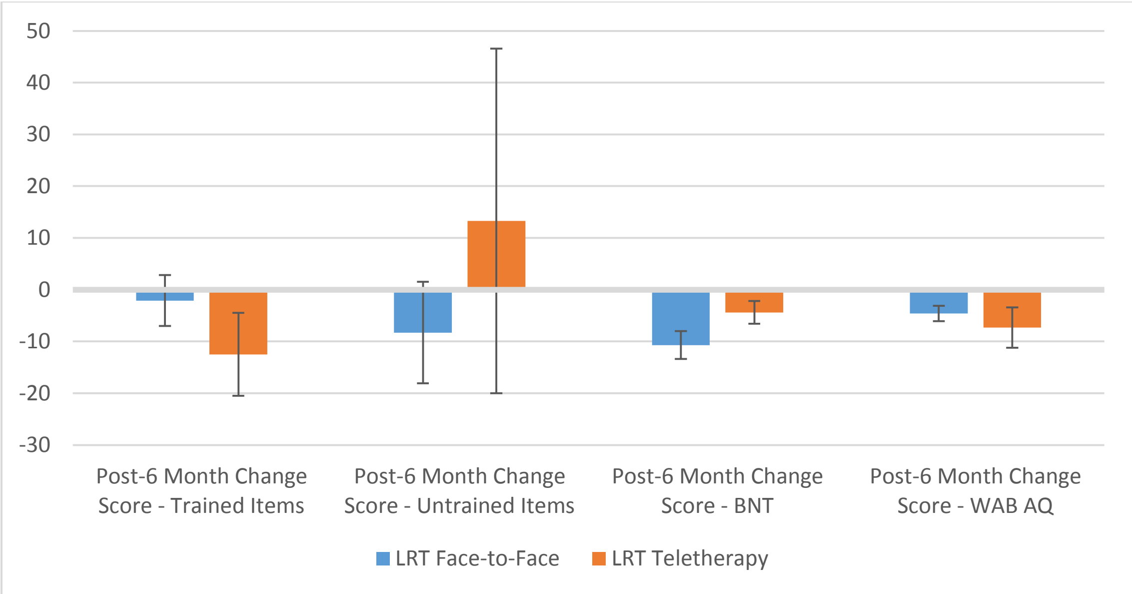
Lastly, maintenance change scores from post-treatment to six months follow-up were also compared between the groups. See Table 8 and Figure 7 for mean change scores from post-treatment to six month follow-up by treatment type. Post-treatment to six month follow-up change scores for trained items for LRT face-to-face (mean rank = 4.33) and LRT teletherapy (mean rank = 6.33) were not significantly different (U = 5.0, z = -1.042, p = .381). Similarly, post-treatment to six month follow-up change scores for untrained items for LRT face-to-face (mean rank = 5.0) and LRT teletherapy (mean rank = 5.0) were not significantly different (U = 9.0, z = .000, p = 1.000). Maintenance on both the *BNT* and the *WAB AQ* was also comparable between LRT face-to-face (mean rank = 5.50, 4.50 respectively) and LRT teletherapy (mean rank = 2.83, 6.00 respectively), as neither the post-treatment to six month follow-up change score for the *BNT* (U = 2.5, z = -1.537, p = .143) or the *WAB AQ* (U = 6.0, z = -.775, p = .548) were significantly different between the two groups.

Figure 6. LRT Face-to-Face Post-3 Month Change Scores vs. LRT Teletherapy Post-3 Month Change Scores. Error bars represent standard errors.



* = $p < 0.05$

Figure 7. LRT Face-to-Face Post-6 Month Change Scores vs. LRT Teletherapy Post-6 Month Change Scores. Error bars represent standard errors.



* = $p < 0.05$

3.2 STATISTICAL ANALYSIS OF VISTA DATA

3.2.1 Treatment Effect and Generalization Following VISTA

Group mean performance on all pre- and post-treatment measures is presented in Table 9 and Figure 8. All test statistics reported for measuring treatment effects and generalization were obtained from Wilcoxon signed ranks tests using a 1-tailed test. All nine participants saw increased percent correct intelligible words in trained scripts following VISTA, with six participants also seeing increased percent correct intelligible words in untrained scripts. This change constituted a significant treatment effect for trained scripts (median change of +57.90%; z = -2.666, p = .004) and untrained scripts (median change of +9.08%; z = -1.820, p = .035).

In examining generalization to standardized tests following VISTA, seven of the eight participants for which a *NAT* score was available saw improvement in their scores following treatment. This change in performance following VISTA was also found to be significant (median change of +18.33%; z = -2.240, p = .013). Similarly, a majority of participants (6/9) improved on their *WAB AQ*; however, this change was not significant (median change of +.60 points; z = -1.362, p = .087).

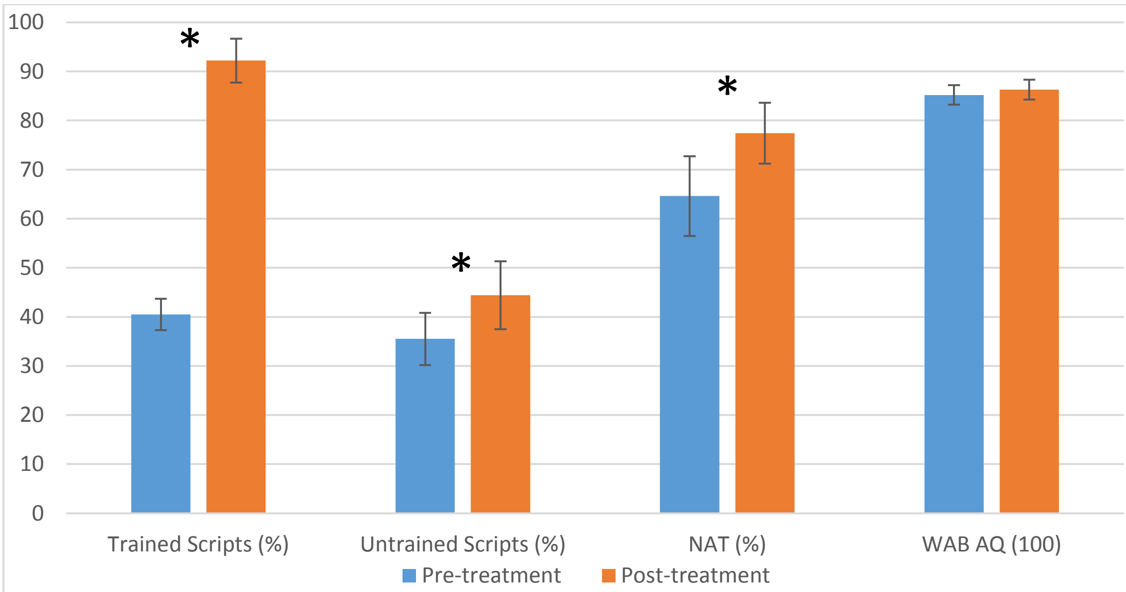
Table 9. Mean Performance on all Dependent Variables (with standard deviations) for All VISTA Participants

	Pre-treatment (*n* = 9)	Post-treatment (*n* = 9)	Post-treatment Equivalent [a] (*n* = 9)	3 Month Follow-up (*n* = 9)	6 Month Follow-up (*n* = 8)
Trained Scripts (%)	40.5 (2.1)	92.2 (13.6)	--	90.8 (17.3)	82.9 (27.7)
Untrained Scripts (%)	35.5 (15.9)	44.4 (20.6)	--	39.9 (19.3)	33.1 (23.1)
NAT (%)	64.6 (22.9) (*n* = 8)[b]	77.4 (18.5)	76.3 (19.1)	69.3 (27.2)	53.8 (35.9) (*n* = 7)[b]
WAB AQ (100)	85.2 (6.0)	86.3 (6.1)	--	82.3 (8.6)	80.1 (9.6)

[a] The full NAT (25-items) was administered at pre- and post-treatment but a shortened version (12 items) was administered at 3 and 6 month follow-up. For each participant with follow-up data, a Post-equivalent *NAT* was calculated by reviewing the scoring protocol and calculating the score on the same set of items, to ensure for valid comparison across time points.
[b] Due to missing data

NAT = Northwestern Anagram Test; *WAB AQ* = Western Aphasia Battery Aphasia Quotient

Figure 8. VISTA Group Mean Performance on all Pre- and Post-treatment Measures. Error bars represent standard errors.



* = $p < 0.05$

3.2.2 Maintenance of Gains Following VISTA

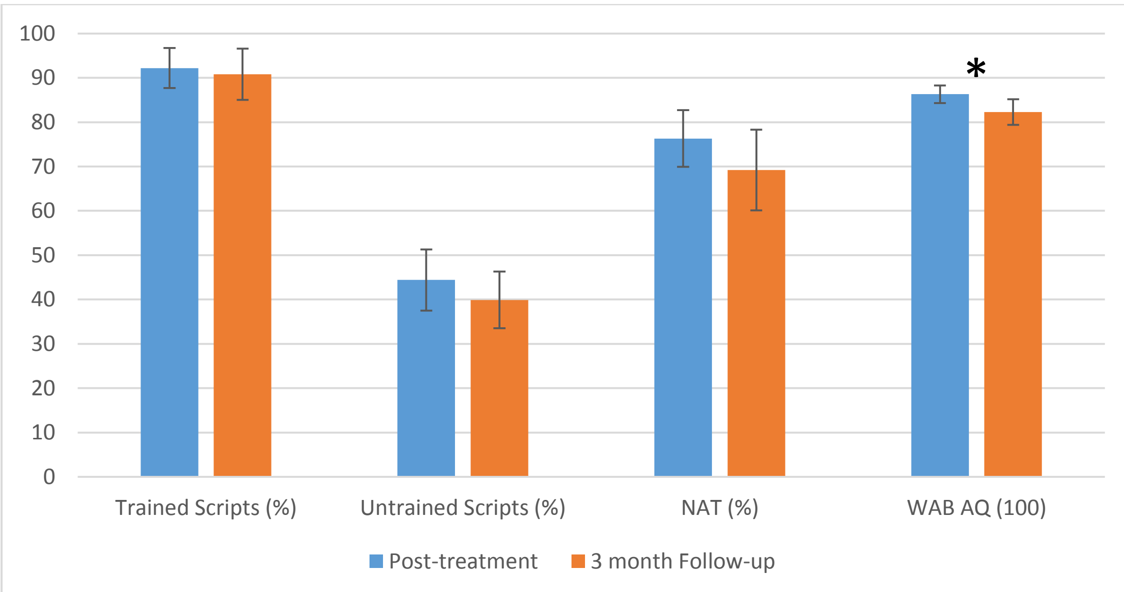
Group mean performance on all post-treatment, three month follow-up, and six month follow-up are presented in Table 9 and Figures 9 and 10. All test statistics reported for measuring maintenance of gains were obtained from a Wilcoxon signed ranks test using a 2-tailed test. As all nine participants had completed three month follow-up testing, all nine were included in the analysis of maintenance of gains from post-treatment to three month follow-up. During this time five of the nine participants experienced declines in percent correct intelligible words in trained scripts. However, this decline was not significant (median change of -.40%; $z = -.652$, $p = .515$). Six of the nine participants also demonstrated decreased performance on untrained scripts at three months follow-up testing. Similar to trained scripts, no significant decline was found on untrained scripts (median change in percent correct of -3.99%; z = -1.125, p = .260), with gains being maintained at three months follow-up.

Maintenance of gains on standardized tests at three months was also examined for both the *NAT* and *WAB AQ*. At three months follow-up, the majority of participants showed either a decline (4/9) or no change (3/9) in their NAT score. Overall, these changes were not significant (median change of .00%; $z = -.943$, $p = .345$). However, significant decline was found on the *WAB AQ* (median change of -2.90 points; $z = -2.666$, $p = .008$), with all nine participants scoring lower at three months follow-up than at post-treatment.

Eight of the original nine participants were also included in the analysis of maintenance from post-treatment to six month follow-up testing. Nearly all participants demonstrated decreased percent correct intelligible words on both trained and untrained scripts (6/8 and 7/8 participants respectively). However, decline on trained scripts was not significant (median change of -2.45%; $z = -1.540$, $p = .123$) whereas decline on untrained scripts was significant (median change of -11.72; $z = -2.380$, $p = .017$) from post-treatment to six months follow-up.

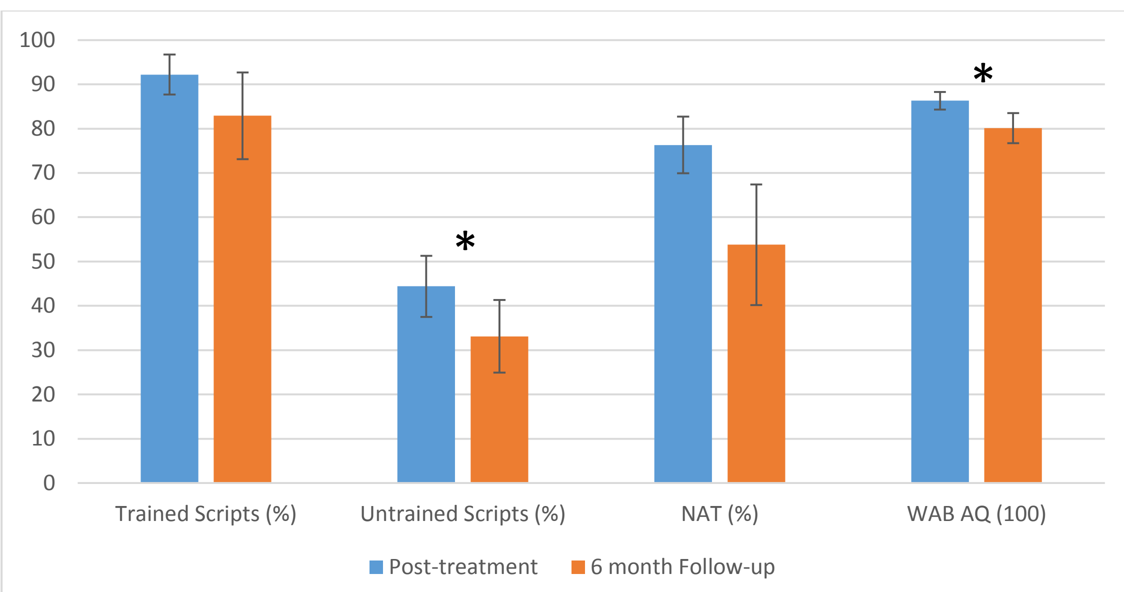
In examining maintenance of performance at six months on standardized tests following VISTA, results were mixed. Though performance on the *NAT* decreased for four of seven participants (only seven scores were available due to missing data), decline at the group level was still not significant (median change of -33.33%; $z = -1.761$, $p = .078$). Conversely, deterioration of scores on the *WAB AQ* was significant at six months follow-up (median change of -4.7 points; $z = -2.028$, $p = .043$), with almost all participants demonstrating a decrease in score (6/8).

Figure 9. VISTA Group Mean Performance on Post-treatment and 3 Month Follow-up Measures. Error bars represent standard errors.



* = $p < 0.05$

Figure 10. VISTA Group Mean Performance on Post-treatment and 6 Month Follow-up Measures. Error bars represent standard errors.



* = $p < 0.05$

3.2.3 Treatment Effect in Traditional Face-to-Face Therapy versus Teletherapy in VISTA Participants

After examining all VISTA cases together for treatment effects and maintenance of gains over time, data were analyzed for between group differences in magnitude of change observed in VISTA face-to-face cases versus VISTA teletherapy cases. Mean percent change scores by treatment type on all pre- and post-treatment measures is presented in Table 10 and Figure 11. All test statistics reported for measuring between group differences were obtained from a Mann-Whitney U test using an exact sampling distribution for U (Dineen & Blakesley, 1973). Demographic variables were also compared between the treatment types. The two groups were not significantly different in age ($U = 5.5$, $z = -1.112$, $p = .286$), education ($U = 9.0$, $z = -.254$, $p = .905$) or pre-treatment *MMSE* score ($U = 4.0$, $z = -1.488$, $p = .190$).

Immediately following treatment, pre-post change scores for trained scripts for VISTA face-to-face (mean rank = 5.00) and VISTA teletherapy (mean rank = 5.00) were not significantly different ($U = 10.0$, $z = .000$, $p = 1.000$). Similarly, pre-post change scores for untrained scripts for VISTA face-to-face (mean rank = 5.40) and VISTA teletherapy (mean rank = 4.50) were not significantly different ($U = 8.0$, $z = -.490$, $p = .730$). Generalization to both the *NAT* and the *WAB AQ* was also comparable between VISTA face-to-face (mean rank = 4.0, 5.2 respectively) and VISTA teletherapy (mean rank = 5.0, 4.75 respectively), as neither the pre-post *NAT* change score ($U = 6.0$, $z = -.577$, $p = .686$) or the pre-post *WAB AQ* change score ($U = 9.0$, $z = -.245$, $p = .905$) were significantly different between the two groups.

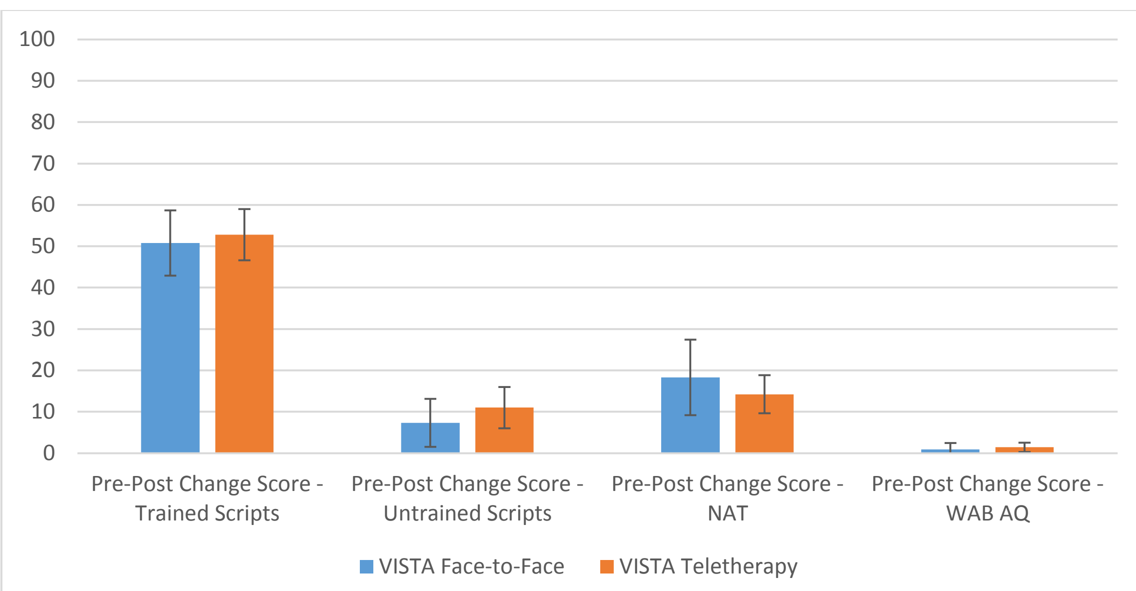
Table 10. Mean Change Scores (with standard deviations) for VISTA Face-to-Face and VISTA Teletherapy.

	Pre-Post Change Score (%)		**Post-3 Month Change Score (%)**		**Post-6 Month Change Score (%)**	
	VISTA Face-to-Face (*n* = 5)	VISTA Teletherapy (*n* = 4)	VISTA Face-to-Face (*n* = 5)	VISTA Teletherapy (*n* = 4)	VISTA Face-to-Face (*n* = 5)	VISTA Teletherapy (*n* = 3)
Trained Scripts	50.8 (17.7)	52.8 (12.3)	-3.2 (6.3)	.79 (2.0)	-14.4 (18.6)	-.55 (1.4)
Untrained Scripts	7.3 (12.9)	11.0 (9.9)	-8.9 (10.3)	.91 (5.7)	-16.6 (11.9)	-7.8 (9.6)
NAT	18.3 (18.1) (*n* = 4)[a]	14.2 (4.6)	-6.0 (28.3)	-8.3 (28.9)	-31.7 (24.6) (*n* = 4)[a]	-13.9 (31.5)
WAB AQ	.86 (3.5)	1.4 (2.2)	-5.3 (5.3)	-2.4 (2.3)	-7.1 (7.0)	-3.7 (7.2)

[a] Due to missing data

NAT = Northwestern Anagram Test; *WAB AQ* = Western Aphasia Battery Aphasia Quotient

Figure 11. VISTA Face-to-Face Pre-Post Change Scores vs. VISTA Teletherapy Pre-Post Change Scores. Error bars represent standard errors.



* = $p < 0.05$

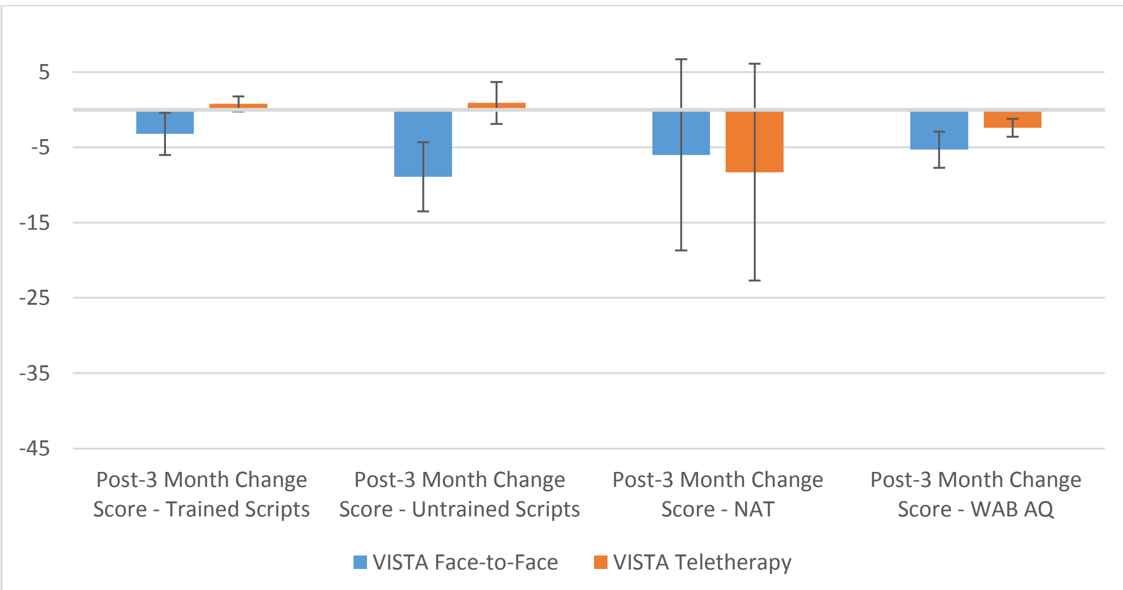
3.2.4 Maintenance Following Traditional Face-to-Face Therapy versus Teletherapy in VISTA Participants

Maintenance change scores from post-treatment to three months follow-up were also compared between the two treatment types. See Table 10 and Figure 12 for mean change scores from post-treatment to three month follow-up by treatment type. Post-treatment to three month follow-up change scores for trained scripts for VISTA face-to-face (mean rank = 5.60) and VISTA teletherapy (mean rank = 4.25) were not significantly different (U = 7.0, z = -.735, p = .556). Similarly, post-treatment to three month follow-up change scores for untrained scripts for VISTA face-to-face (mean rank = 6.20) and VISTA teletherapy (mean rank = 3.50) were not significantly different (U = 4.0, z = -1,470, p = .190). Maintenance on both the *NAT* and the *WAB AQ* was also comparable between VISTA face-to-face (mean rank = 5.20, 5.40 respectively) and VISTA teletherapy (mean rank = 4.75, 4.50 respectively), as neither the post-treatment to three month follow-up change score for the *NAT* (U = 9.0, z = -.249, p = .905) or the *WAB AQ* (U = 8.0, z = -.490, p = .730) were significantly different between the two groups.

Lastly, maintenance change scores from post-treatment to six months follow-up were also compared between the groups. See Table 10 and Figure 13 for mean change scores from post-treatment to six month follow-up by treatment type. Post-treatment to six month follow-up change scores for trained scripts for VISTA face-to-face (mean rank = 5.40) and VISTA teletherapy (mean rank = 3.00) ad were not significantly different (U = 3.0, z = -1.342, p = .250). Similarly, post-treatment to six month follow-up change scores for untrained scripts for VISTA face-to-face (mean rank = 5.00) and VISTA teletherapy (mean rank = 3.67) were not significantly different (U = 5.0, z = -.745, p = .571). Maintenance on both the *NAT* and the *WAB AQ* was also comparable between

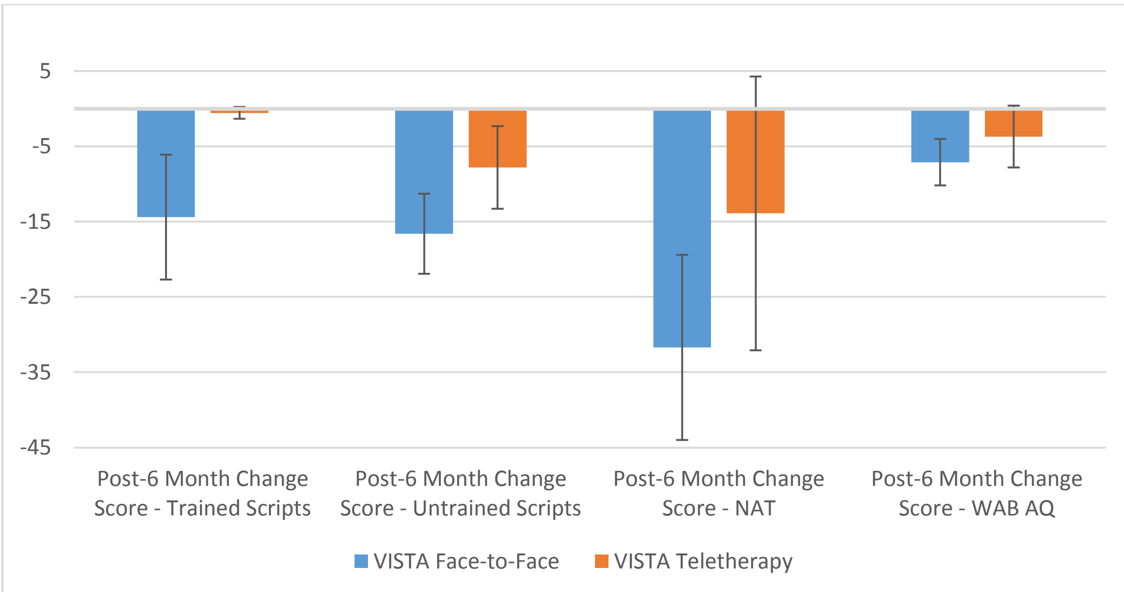
VISTA face-to-face (mean rank = 4.63, 5.20 respectively) and VISTA teletherapy (mean rank = 3.17, 3.33 respectively), as neither the post-treatment to six month follow-up change score for the *NAT* (U = 3.5, z = -.900, p = .400) or the *WAB AQ* (U = 4.0, z = -1.043, p = .393) were significantly different between the two groups.

Figure 12. VISTA Face-to-Face Post-3 Month Change Scores vs. VISTA Teletherapy Post-3 Month Change Scores. Error bars represent standard errors.



* = $p < 0.05$

Figure 13. VISTA Face-to-Face Post-6 Month Change Scores vs. VISTA Teletherapy Post-6 Month Change Scores. Error bars represent standard errors.



* = $p < 0.05$

4. Discussion

To date, almost no studies exist that examine treatment outcomes in traditional face-to-face therapy versus teletherapy in individuals with PPA. In the current study, we have presented data exploring the efficacy of teletherapy for treatment of individuals with PPA using a lexical retrieval protocol and a script training protocol. A total of 25 participants were assigned to one of the two treatment protocols, with protocol assignment based on the core linguistic deficits associated with their PPA subtype. Lexical Retrieval Cascade Treatment (LRT), the treatment approach implemented with individuals with semantic or logopenic variant of PPA, utilized a cueing hierarchy designed to activate residual semantic, orthographic, and phonological information to facilitate naming abilities. Video-Implemented Script Training in Aphasia (VISTA), the treatment approach implemented with individuals with non-fluent/agrammatic variant PPA, was designed to improve speech production, intelligibility, and fluency for functional, scripted material. Within each treatment protocol, one group of participants received treatment via teletherapy, while the other group received treatment via traditional face-to-face treatment sessions.

Treatment outcomes indicated that both LRT and VISTA protocols resulted in a significant treatment effect for trained items, as all participants significantly improved either their ability to consistently name a set of nouns or fluently and intelligibly produce a set of personalized scripts. Participants receiving LRT also showed generalization of the retrieval strategy to untrained nouns and the *BNT*, a standardized test of naming ability. In addition, LRT participants demonstrated maintenance of gains on both trained and untrained items up to six months post-treatment, as well as maintenance on the *BNT* up to three months post-treatment. Participant receiving VISTA also demonstrated generalization and maintenance of gains following treatment, although in a slightly different pattern than LRT participants. VISTA participants also showed generalization to untrained scripts, as well as to the *NAT*, an assessment of syntactic production. Following VISTA, gains on both trained scripts and the *NAT* were maintained up to six months post-treatment, while gains on untrained scripts were maintained only at three months post-treatment. Thus, both treatments had a selective treatment effect on the linguistic parameters targeted by the protocol (i.e. word retrieval in LRT and syntax production in VISTA) that generalized to assessments of these parameters. These lasting and generalized effects following treatment were observed in the context of inevitable language deterioration, as measured by the *WAB AQ*, a standardized assessment spanning several linguistic domains. Neither group of participants demonstrated generalized improvement on the *WAB AQ* following treatment. Furthermore, both LRT and VISTA participants experienced significant decline on the *WAB AQ* at both three months and six months post-treatment.

While both LRT and VISTA protocols resulted in significant treatment effects on trained and untrained variables, our primary question was whether or not the magnitude of change following treatment was comparable between teletherapy and face-to-face participants in both groups. Current research literature in stroke-induced aphasia has shown that treatment outcomes between teletherapy and traditional face-to-face therapy are equivalent (Cherney & Van Vuuren, 2012; Hall, Boisvert, & Steele, 2013). Given these findings, we predicted that treatment outcomes between teletherapy and face-to-face therapy would also be comparable in PPA. As predicted, analyses revealed that, regardless of treatment approach (LRT or VISTA), treatment effects and generalization to untrained items and standardized tests in participants receiving teletherapy were comparable to those seen in participants receiving traditional therapy. Likewise, observed maintenance of gains or decline in abilities over time was comparable between teletherapy and face-to-face participants in both treatment protocols. These findings indicate that teletherapy is an effective method for delivering intervention for individuals in PPA, as it offers treatment benefits comparable to those seen in face-to-face therapy.

A strength of this study was the validation of teletherapy as a valuable tool in delivering treatment to a patient population that is underserved due to concerns about utility of treatment in progressive diseases and issues in accessing appropriate treatment. Thus far, teletherapy has not been thoroughly explored in patients with this clinical diagnosis, despite related work in stroke-induced aphasia demonstrating the feasibility of teletherapy in addressing neurogenic language disorders. Furthermore, this study explored teletherapy outcomes in a treatment protocol targeting linguistic skills other than lexical retrieval, which to date has been the main focus in both face-to-face PPA treatment studies and the focus of the only published PPA teletherapy study. The study did, however, have some limitations. Although comparable treatment effects, generalization, and maintenance were observed in teletherapy and traditional therapy, this study had a small number of participants. Further, participants were not randomly selected to receive therapy via video-conferencing or in person; instead participant need dictated group membership, just as would occur in actual clinical practice. Additionally, this study only utilized two treatment approaches, LRT and VISTA, both of which were easily adapted for and implemented via video-conferencing software. Other treatment approaches may not be comparable across teletherapy and face-to-face therapy due to limitations in adapting an approach for delivery via video-conferencing or the nature of linguistic characteristics targeted by the protocol. Lastly, an important consideration is that our study only included participants with mild to moderate cognitive and linguistic deficits, and thus we are unable to comment on the utility of teletherapy with more moderate-severe to severe patients. Teletherapy benefits in more severe patients may be more limited given the increased technical and linguistic demands imposed by this medium. In particular, the combination of auditory comprehension deficits and the degraded auditory-visual signal present in most video-conferencing technologies may prove challenging for more severely impaired patients. However, benefits are not constrained by PPA subtype, as we have shown that the three clinical variants can benefit equally. While patient subtype was not an explicit variable analyzed in our data, teletherapy and face-to-face treatment groups included participants of all three variants, suggesting that all variants benefitted from both forms of treatment delivery.

Future research is needed to explore the inclusion of patients with severe deficits in treatments administered by teleconference. This could include investigating the development of treatment approaches targeting more relevant variables for severe patients (e.g. use of alternative or augmentative communication) and comparing outcomes to those seen in traditional therapy. Further, future studies can address what cognitive and/or linguistic deficits in moderate-severe to severe patients may predict what, if any, benefit a patient may obtain from treatment administered via teletherapy. Additional directions for future research include broadening the sample size and/or including participants with other etiologies. Replication of this study in a larger sample of individuals with PPA would allow us to examine whether trends seen in this study are present in a larger group of participants. Analyzing the efficacy of teletherapy in patients with other etiologies, such as mild dementia, traumatic brain injury, or mild cognitive impairment, could lead to even more patient populations being reached that typically receive little-to-no speech-language therapy.

In summary, our results support the use teletherapy as an efficacious method of treatment delivery for individuals with PPA. Teletherapy has already begun to emerge as a viable option for treatment in various speech and language disorders, such as stroke-induced aphasia, as well as in other fields of rehabilitation (e.g. physical therapy). As the research base continues to establish teletherapy as a viable alternative to traditional therapy, increased provision of services will be possible. The advantages of teletherapy in mitigating issues of treatment access will prove especially beneficial in populations that have been historically underserved as a result of distance, mobility issues, or unavailability of specialists.

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