EFFECTS OF MODIFIED VIDEO IMPLEMENTED SCRIPT TRAINING FOR APHASIA IN THE THREE VARIANTS OF PRIMARY PROGRESSIVE APHASIA

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ABSTRACT

PURPOSE: Primary progressive aphasia (PPA) is a neurodegenerative disorder characterized by worsening of speech and/or language. Script training intervention promotes automatized speech production via repeated practice of scripted content. This study evaluated the acceptability, feasibility and effects of a modified version of Video Implemented Script Training for Aphasia (VISTA) in the three PPA variants and compared outcomes by intervention modality (teletherapy versus in-person).

METHODS: Thirteen bilingual (Spanish-Catalan) participants were included (semantic variant, n = 5; logopenic variant, n = 5; nonfluent/agrammatic variant, n = 3; teletherapy, n = 7). Using a non-randomized design, intervention was administered in participants' dominant language. Participants were trained on an individualized script twice per week, over eight-weeks. Performance on measures related to script accuracy, content, and subjective ratings of production quality were evaluated at baseline, immediately post, and at three- and six-months post-intervention.

RESULTS: No significant differences were observed on the basis of intervention modality. Participants demonstrated significant improvements from pre- to post-intervention in script production, synonym production, keywords, and global quality on the trained script. Maintenance was observed when comparing performance at post-intervention relative to three- and six-month follow-up for script and synonym production. Significant improvement in production quality of the untrained topic was observed following intervention. Different patterns of benefit were observed by PPA variant.

DISCUSSION: Modified VISTA was acceptable and effective across the three PPA variants, as evidenced by improvements on a broader array of outcome measures than those previously reported. Findings also provide further support for provision for teletherapy in individuals with PPA.

KEYWORDS: Primary Progressive Aphasia; Spanish-Catalan; bilingual; intervention; script training; teletherapy

1.INTRODUCTION

Primary progressive aphasia (PPA) is a clinical syndrome characterized by progressive worsening of speech and/or language due to an underlying neurodegenerative process, with relative sparing of other cognitive and behavioral abilities (Gorno-Tempini et al., 2011; Mesulam, 1982; Olney et al., 2017). Currently, three PPA variants have been described, each with a different profile of speech and/or language impairment: Semantic variant PPA (svPPA) presents with loss of vocabulary and semantic deficits, including impairment of single-word comprehension (Gil-Navarro et al., 2013; Gorno-Tempini et al., 2011; Marshall et al., 2018). Logopenic variant of PPA (lvPPA) is associated with impaired word retrieval, sentence repetition, and phonological errors (Gil-Navarro et al., 2013; Gorno-Tempini et al., 2011). Nonfluent/agrammatic variant PPA (nfvPPA) presents with apraxia of speech and/or agrammatism (Croot et al., 2012; De Leon et al., 2019; Gil-Navarro et al., 2013; Gorno-Tempini et al., 2011). A minority of people present with mixed features, thus leading to the categorization of these individuals as having "unclassifiable" PPA (Gorno-Tempini et al., 2011; Harris et al., 2013; Montembeault et al., 2018).

Research studies on the effects of behavioral speech-language intervention in PPA have increased in the last couple of decades (Croot et al., 2009; Jokel et al., 2014; Kortte & Rogalski, 2013; Rising, 2014; Tippett et al., 2015; Wauters et al., 2023). This increase has led to the identification and implementation of several interventions and compensatory strategies that have proven to be successful with people with PPA (Pagnoni et al., 2021). However, these efforts still remain sparse compared with post-stroke aphasia.

In stroke-induced aphasia, an intervention approach broadly known as script training, focuses on the repeated practice of scripted content to promote automated and fluent speech production. Script training has a long history in the stroke-induced aphasia literature (Ali et al., 2018; Cherney et al., 2008; Cherney, 2012; Goldberg et al., 2012; Grasso et al., 2019; Holland et

al., 2002; Hubbard et al., 2020; Rhodes & Isaki, 2018) and has been shown to have a positive effect on encoding, retrieval, and accurate production of scripted content (i.e., percentage of accurately produced scripted words) as well as on speech rate (i.e., number of script related words produced per minute; (Cherney, 2012). The theoretical underpinnings of script training are derived from the instance theory of automatization, which suggests that the automaticity of instances or episodes of learning is achieved by retrieving memories linked to a context (Logan, 1988). The goal of script training is to achieve fluent, automatic speech by repeatedly practicing scripted content for use in daily life. To ensure functionality, scripted content should be relevant to peoples' daily lives and communicative needs. In this sense, the personalization of scripted content is considered critical for engagement in learning scripted content (Cherney et al., 2015).

Computer- and avatar-based administrations of script training have been previously reported in stroke-induced aphasia (Bilda, 2011; Cherney et al., 2007; Cherney et al., 2008), resulting in clear communicative improvements (e.g., scripted word accuracy, rate of production, grammatical productivity). The approach has also been proven to be effective via tele-based intervention (Hubbard et al., 2020; Rhodes & Isaki, 2018). In contrast to the robust literature investigating script training in stroke-induced aphasia, only a few studies have evaluated the effects of this approach in individuals with PPA. An approach to script training developed by Henry et al., 2018, provided evidence for the positive effects of Video-Implemented Script Training for Aphasia (VISTA) in individuals with nfvPPA, specifically. In this approach, individuals are provided with personalized videos/scripts that they practice speaking in unison with daily, coupled with twice weekly sessions with clinicians to practice memorization and conversational usage of scripted content (Fridriksson et al., 2012). Of note, no significant differences have been found in individuals with mild to moderate nfvPPA who were administered VISTA via teletherapy vs. in-person (Dial et al., 2019). In nfvPPA, VISTA has also been shown to be effective when modified to account for hearing loss, wherein the original VISTA treatment was modified by providing practice with orthographic input as a supplement to

audiovisual input during script practice (Schaffer et al., 2022). Additionally, VISTA has been used in conjunction with aphasia-modified cognitive behavioral therapy in the context of nfvPPA (Schaffer et al., 2021), which demonstrated improvements in script accuracy and psychosocial and communicative functioning. Although the effects of script training in nfvPPA have been previously reported, the effects of script training have yet to be examined in the logopenic or semantic variants of PPA.

In the current study, we hypothesized that script training would result in improved communication among the three variants of PPA, given that this training is person-centered and addresses several elements of speech and language production (e.g., fluency, word finding, and selection). In nfvPPA, script training addresses the core communication deficits in speech production and agrammatism. However, individuals with lvPPA also struggle with fluent production, which is largely driven by pauses for word retrieval (Gorno-Tempini et al., 2011). Therefore, we hypothesized that script training may prove useful in lvPPA by providing rehearsed practice with established content, which may lower the demands on word finding. Individuals with svPPA present with fluent speech production, yet often produce empty language (Gorno-Tempini et al., 2011), as such script training may benefit these individuals by improving the specificity and meaningfulness of their output.

Therefore, consistent with the NIH stage model (Onken et al., 2014), we conducted a pilot study to evaluate the feasibility and acceptability, and intervention effects of a modified version of VISTA (Henry et al., 2018) in bilingual (Spanish-Catalan) participants with the three PPA variants. We also compared the effects of a modified version of VISTA administered via two modalities: in-person intervention versus teletherapy (specific details of how VISTA was modified can be found in section 2.3.3). We predicted that modified VISTA would result in improvements at the group level and that there would be no significant differences based on intervention modality. Lastly, we hypothesized that patterns of intervention response could vary

in different PPA variants and therefore conducted an initial characterization of variant-specific effects following the administration of modified VISTA.

2. METHOD

2.1. Design

The current study design was consistent with the NIH Stage Model Stage 1B classification (Onken et al., 2014), a framework that reflects stages of intervention development in a clinical science research. A non-randomized quasi-experimental design was used. Participants were assigned to in-person versus teletherapy intervention according to each participant's geographical (distance of residence from the hospital), physical (mobility issues), and technical (internet access, computer, or tablet) conditions. To qualify for teletherapy, participants needed to meet at least one of the following criteria: living outside of Barcelona, experiencing difficulties with walking and therefore at risk of falling, or if they indicated a strong preference for this modality.

2.2. Participants

To be eligible for enrollment in the study, participants were required to meet the current consensus criteria for one of the three PPA variants (Gorno-Tempini et al., 2011). Thirteen individuals, three men and ten women, with mild-to-moderate PPA participated in the study: five with svPPA, five with lvPPA, and three with nfvPPA. All three participants with nfvPPA presented with clinical symptoms of apraxia of speech, two had dysarthria and none of them were frankly agrammatic. The mean age of the participants was 70.7 years (SD = 5.6) and the mean years of education were 11.4 years (SD = 4.1). Six of them received in-person intervention and seven received teletherapy. All participants were bilingual (Spanish-Catalan), though intervention was administered in participants' dominant language. Diagnoses by variant were performed by a behavioral neurologist and supported by the neuroimaging findings through structural magnetic resonance imaging and/or PET-FDG. Additional criteria included a Mini-

Mental State Examination (MMSE) (Folstein et al., 1975) score of 15 or higher. This cut-off was based upon previous studies utilizing VISTA which employed the same cut-off (Dial et al., 2019; Henry et al., 2019). Participants were also required to commit to attend speech and language therapy sessions twice a week during the study period. Beyond the aforementioned criteria surrounding teletherapy, to be eligible for teletherapy, participants were required to have computer or tablet and Internet access, a study partner who could provide technology support (if needed), and an absence of uncorrected vision or hearing impairments.

Consecutive participants meeting the inclusion criteria were offered to participate in the study at the Alzheimer's and Other Cognitive Disorders Unit of the Hospital Clínic de Barcelona (Barcelona, Spain). All participants gave written informed consent to participate. Supplementary verbal explanations were offered to those who had difficulty understanding relevant written terms, as well as to their primary caregivers. This study was approved by the Hospital Clínic de Barcelona Ethics Committee (HCB/2019/0985). See Table 1 for the participants' demographic information.

2.3. Procedures

2.3.1 Cognitive and linguistic assessment

The participants underwent a neurological and general cognitive evaluation at baseline by a neurologist and neuropsychologist. Linguistic and speech examinations were performed at baseline and at each time point by a speech-language pathologist (SLP). To evaluate general cognitive status, we used the MMSE. Confrontation naming and cognitive abilities were assessed using the naming section of the Cambridge Semantic Battery (Bozeat et al., 2000), and Digit Span (forward and backward) (Wechsler, 1997). To evaluate motor speech and non-speech abilities, we administered a quantitative assessment tool, The Barcelona Scale for Buccophonatory

Apraxia (Montagut et al., 2022), see Table 1. Briefly, the BSBA is a measure that was developed to indicate the optimal cut-off point (215 seconds; see Montagut et al., 2022) for differentiating individuals with nfvPPA with apraxia of speech (AOS) from other syndromes. A greater duration in seconds is generally indicative of greater impairment in motor speech and non-speech abilities.

2.3.2. Script development

An individualized script entitled "My Story" was created for each participant. The script contained three sentences related to each participant's daily life. These sentences were developed in response to the following three questions: 1: "Do you have any difficulties?" 2: "What was your job?" and 3: "What do you like to do now?". Participant responses were audio recorded by the SLP. The SLP then created a personalized script with the participants and their caregivers to ensure the reliability of their responses. If the participant produced sufficient content in response to the topic spontaneously, the SLP utilized solely this information provided by the patient to create the script. The script was then sent via email to the patient and caregiver for their review and any necessary modifications were made by the SLP (e.g., accuracy of content). If the participant's spontaneous production yielded insufficient information for the SLP to develop the script, the SLP contacted the caregiver to acquire the necessary details to construct the script. Then, the process was conducted as described in the first scenario (i.e., the SLP created the script which was sent to the patient and caregiver for approval, and their suggested modifications were incorporated; see Supplementary Material for an example script).

The final script had a similar number of words as the participants' spontaneous responses to the clinician-led questions. If a participant's spontaneous response was limited, the SLP added the minimum number of words necessary to create a reasonable and functional script. The rationale for this individualized "My Story" script was to provide participants with responses to frequently asked questions, thereby automating their typical responses in daily

communication. Each participant's script was developed so that they could respond to individual sentences from their script or with the entire script as a unit. Performance on an untrained and unscripted topic (i.e., no scripted target was established), was assessed by asking the same question at the end of each session ('What will you do today?').

2.3.3. Script training

All methods described herein were identical for participants who received intervention in-person and via teletherapy. The only difference is that these procedures were conducted either via tele-based means in a synchronous fashion or in-person. Individuals received a modified version of VISTA twice per week (30 min per session) over an eight-week period. The intervention was administered synchronously always with the same bilingual speech and language pathologist (Núria Montagut) who has extensive experience with administering speech-language intervention to individuals with neurodegenerative diseases. Intervention and script videos were administered in each participant's self-reported dominant language (Spanish or Catalan), to ensure functionality. Relative to the original version of VISTA (Table 2), the modified version of VISTA used in the current study contained one trained script instead of four, sessions were shorter in duration (30 min instead of 45 min to 1 h), our overall intervention period was longer (8 weeks vs. 4-6 weeks), and our untrained topic was not scripted (whereas the original approach included two untrained scripts). All participants were trained on individualized scripts.

A seven-step hierarchy was used to target the accuracy of production and conversational usage of scripted content (Table 2). In addition to memorization of scripted content, we focused on articulatory and/or phonological aspects and supported participants' accurate production of scripted content by providing them with cues (i.e., instructing individuals to articulate the difficult word with a pause between each syllable, imitating the exaggerated articulation from the SLP's example). These cues are used when an individual demonstrates

motor speech or phonological errors. For individuals who demonstrated phonological errors (those with lvPPA), the first syllable was also provided as a cue if the individual demonstrated difficulty reading or producing the word. Individuals with svPPA were asked to provide the meaning of the content words embedded in their script during steps that required reading or spontaneous production of the script. For both lvPPA and svPPA, if word retrieval was unsuccessful after being provided cues, a personalized and functional cloze phrase was provided to elicit the word ("I sit on the sofa after lunch to watch _____", for the target word "TV"). Finally, individuals with svPPA were asked to affirm or deny their understanding of words in their scripts. If they did not understand the sentences in their script, the clinician explained the content.

Videos were created for the personalized script by recording the mouth of a healthy speaker, producing the script at a speech rate approximating that of the participant. These videos were used for independent home practice without SLP. The practice consisted of unison speech production with the audio-visual model three days per week (a single production was requested during each practice session). Lastly, it was recommended that participants take advantage of communicative situations with other people by using the trained script when appropriate. Participants did not receive any other type of speech-language intervention outside of the present study from pre- to post-intervention. Because these participants were recruited via the hospital where the first author is employed, participants were invited to continue with their regular group therapy sessions after post-intervention testing.

With respect to intervention administration, the first author developed a manual for the implementation of the intervention that included the hierarchy of steps to be followed for each patient, during each session. These steps are summarized in Table 2. Before each session, the clinician reviewed the written hierarchy of steps and had the written steps available during the intervention to ensure high fidelity of administration. Lastly, for participants who received teletherapy, no technical or connectivity issues arose.

2.4. Outcome Measures

For the trained script, we investigated changes in the following outcome measures in the entire cohort of participants: script production, keyword production, synonym production, and the overall global quality of script production. For the untrained and unscripted topics, we only evaluated overall global quality, as an individual's untrained topic did not have a predetermined scripted target. Secondary outcome measures included performance on selected assessment measures collected at baseline and longitudinally (naming task from the Cambridge Semantic Battery and the MMSE). The potential for generalization to untrained and unscripted topics was assessed via global quality and a naming task from the Cambridge Semantic Battery. Performance on outcome measures was assessed using audio recordings of participants' responses at baseline, post-intervention (within one week after therapy ended), and at three- and six-months after intervention. The baseline evaluation was undertaken by the first author. A second SLP (Jorge Herrero), a naïve listener, who was blinded to the time point of each recording and was not involved in the administration of therapy, evaluated the performance of participants on all outcome measures.

Script Production: Script production is defined as the number of correct and intelligible words produced by the individual relative to the scripted target. Phonological and semantic paraphasias were not counted as correct in the total score. The proportion of correct and intelligible scripted words was calculated using a denominator that represented the total number of words present in the script. Some iteration of this outcome measure has been utilized in most of the previous work investigating the effects of script training in order to assess overall learning of the scripted content (Hubbard et al., 2020).

Keyword Production: Keyword production was defined as the sum of correct nouns, verbs, adjectives, and adverbs relative to the total number of keywords present in the script. The proportion of keywords produced was calculated using a denominator that represented the

total number of *keywords* present in the script. If keywords were distorted or were produced with a phonological paraphasia, but were still intelligible, they were counted as correct. Completely unintelligible words were not counted as correct. We selected this measure because we predicted that the keywords would have the greatest semantic weight when producing scripted content.

Synonyms: Synonym production is defined as the number of synonyms produced in place of the keywords present in the script. The proportion of synonyms produced was calculated using a denominator that represented the total number of *keywords* present in the script. We included this measure because we anticipated that individuals with phonological processing impairment (lvPPA) and conceptual impairment (svPPA) may use synonyms instead of scripted keywords and therefore sought to capture potential improvements in synonym production.

Global Quality: Global quality is defined as the qualitative rating of the production quality. Ratings were related to intelligibility/pronunciation (i.e., participant's ability to produce the words clearly), fluency (i.e., participant's continuous and fluent production of words and connected speech), vocabulary (i.e., participant's correct use of words to express what is meant), and overall script coherence (i.e., participant's ability to express ideas in an organized and logically connected way), with the assignment of an overall qualitative score corresponding to severe (1), moderate to severe (1.5), moderate (2), mild-moderate (2.5), mild (3), normal-mild (3.5), and normal (4). We included this measure in order to have a qualitative rating that was taking into account the overall quality of the individual's production.

2.5. Post-intervention Perceived Satisfaction/Acceptability Questionnaire

A five-item Perceived Satisfaction/Acceptability Questionnaire**,** using a Likert-type scale, was administered post-intervention to all participants as a measure of acceptability. Participants were encouraged to complete the survey with their study partners or care partners whenever possible to ensure that they understood the questions. The questions were related to an

individual's perceived satisfaction with therapy. Each item was rated on a five-point scale: 1. Strongly disagree, 2. Disagree, 3. Neither agree nor disagree; 4. Agree, 5. Strongly agree; see Supplementary Material.

2.6. Data preparation and analysis

First, independent-samples permutation tests were conducted between individuals who received in-person intervention and those who received teletherapy. Change scores were calculated at the individual level by subtracting pre-intervention performance from post-intervention performance as well as pre-intervention performance relative to follow-up observations. We evaluated the results of these analyses using two-tailed tests, as we predicted no significant differences based on intervention modality. The statistical outcome measures for the trained script at the group level included script production, keyword production, synonyms, and overall global quality. For the untrained and unscripted topic, only the measure of global quality was assessed.

At the group level, nonparametric exact permutation tests were conducted using the COIN package in R (version 4.2.1, 2022) (Hothorn et al., 2008) or custom scripts. We report the T-values as well as the exact significance levels derived from permutation analyses. At the group level, baseline performance was compared to post-intervention and at the- and six-months follow-up via paired-samples permutation tests. One-tailed tests were used to examine trained script performance for these analyses, as we predicted significant improvement following the intervention and at subsequent follow-ups. Two-tailed tests were used to compare untrained topics from pre- to post-intervention (for global quality) and performance on a confrontation naming test, as generalization effects were less predictable. We also examined the maintenance effects by comparing performance at post-intervention relative to subsequent follow-ups using two-tailed paired permutation tests, with a nonsignificant p-value indicating maintenance.

MMSE performance was also evaluated over time using two-tailed paired permutation tests to assess the potential for changes in cognitive status throughout the study.

We were interested in examining individual responses to intervention given that previous reports have indicated heterogeneity. Significance testing at the individual participant level was conducted using simulation where appropriate (Dial & Martin, 2017) (except for global quality due to the noncontinuous nature of this outcome measure). To do so, each participant's accuracy (expressed as a percentage of the total output) was calculated from each time point for the trained script, which was used to generate probabilities of correct responses to create simulated datasets with parameters that aligned with the observed data. 10,000 simulated distributions of percent accuracy were generated, and the resulting simulated datasets from two time points were then directly compared to calculate the p-value. In addition, using the simulated data, difference scores were calculated between the time points to determine the 95% confidence intervals of the observed differences.

3. RESULTS

3.1. Intervention effect and modality of the intervention

No significant differences were observed in the magnitude of the intervention (Figure 1) or generalization effects for any comparison from pre- to post-intervention or from pre- to each subsequent follow-up between the intervention modalities (in-person vs. teletherapy). We also did not observe any significant differences between modality on other outcome measures (i.e., keywords, synonyms and global quality; all $p > .10$). Given that we did not observe differences between intervention modalities, all subsequent analyses combined participants who received in-person intervention and those who received teletherapy.

3.2. Group Level Analyses

3.2.1. Intervention and Maintenance Effects

For the trained script, we observed a significant improvement from pre- to post-intervention in script production (t = -1.86, p = .040), keywords (t = -5.32, p = .001), synonyms (t = -3.50, p = .004), and global quality (t = -4.45, p = .001) (Figure 2A and 2B). Maintenance was observed when comparing performance at post-intervention relative to the three- and six-month follow-ups for script and synonym production, whereas a lack of maintenance was observed for keyword and global quality at six-months post-intervention. Nevertheless, script production, keywords, synonym production, and global quality were significantly better at both follow-ups than at baseline (all p-values < .05). For the untrained topic, we observed a significant generalization of global quality from pre- to post-intervention (t = -2.13, p = .049), although maintenance was not observed at subsequent follow-ups. (Figure 2A and 2B).

3.2.2. Secondary Outcome Measures

We observed significant generalization effects on the naming task from the Cambridge Semantic Battery post-intervention relative to the baseline (t = -2.45, p = .002). However, this effect was not maintained at the three- or six-month follow-up. Nevertheless, participant performance on the Cambridge Naming Test was maintained at follow-up and did not show decline. Lastly, no significant changes were observed in MMSE scores over time (Figure 3).

3.3. Individual Participant Level Analysis and Patterns by PPA Variant

Table 3 shows the results of the simulation analyses, which revealed that at the individual participant level, one participant demonstrated a statistically significant improvement in script production from pre- to post- intervention (M change = 4%, range = -8-22%). Out of the 13 participants, nine individuals showed significant improvements in keyword production (M change = 41%, range 0-80%), and only one individual showed significant improvement in synonym production (M change = 5%, range 0-13%). Most participants who demonstrated a

significant effect from pre- to post-intervention also showed significant maintenance at the three-month follow-up and some individuals showed this pattern at six-month follow-up (see Table 3).

With respect to patterns by PPA variant, significant improvements in keyword production were observed in lv and nfvPPA. Numerical improvements were also observed for script production, largely in lv and nfvPPA, whereas improvements in synonym production were largely observed in lvPPA. In individuals with svPPA, we observed a ceiling effect in script production and minimal numerical improvement in keyword and synonym production (see Table 3).

3.4. Perceived Satisfaction/Acceptability Questionnaire

All participants completed the five-item satisfaction/acceptability questionnaire. In-person participants completed the post-intervention survey on paper, and for the participants who received teletherapy, we emailed the survey using Google Forms. General satisfaction/acceptability with the intervention was high, with an average of 4.8 out of 5. In addition, participants reported that the therapy benefitted their everyday communication and expressed a desire to continue with the speech-language intervention that was offered. The items that were most closely linked to acceptability were those that queried 1) if therapy benefitted their everyday communication and 2) their overall satisfaction with the intervention. All participants rated these items with a rating of "agree" or "strongly agree". None of the participants selected a rating of “disagree” or “strongly disagree” for any of the survey items. Ratings representing the overall degree of satisfaction with the intervention were comparable between participants who received in-person (average of 4.7 out of 5) and teletherapy (average of 4.9 out of 5), see Figure 4.

4. DISCUSSION

In the present study, we evaluated the effects of a modified version of VISTA in the three variants of PPA. We sought to establish the acceptability and feasibility of this intervention and compare the effects of script training administered via teletherapy versus in-person training. Participants were Spanish-Catalan bilingual speakers, who received intervention in their dominant language, administered by a bilingual/bicultural speech-language pathologist, thereby ensuring the intervention was culturally and linguistically tailored. Our results indicate that the modified version of VISTA utilized herein is an effective intervention approach for individuals with svPPA, lvPPA, and nfvPPA, as evidenced by improvements in a broader array of outcome measures (i.e., keywords, use of synonyms, and global quality) than those previously reported in the literature.

4.1. Contextualizing immediate and maintenance effects

Consistent with our hypothesis, a significant improvement was observed at the group level on all outcome measures from baseline to post-intervention (i.e., script production, keywords, synonyms, and global quality). Maintenance was observed from post- to three- and six-months post-intervention for the production of trained scripts (i.e., correct and intelligible scripted words) and synonym production. Although neither keywords nor global quality showed maintenance at 6-months post-intervention, performance was still better when compared to pre-intervention. Given that individuals continue to decline with progressive worsening of speech and language, this pattern of performance indicates maintenance that is above baseline performance. Maintaining the benefits of the intervention over time can be challenging, particularly due to the progressive nature of the disease and the associated cognitive and language decline. These factors can impact the ability to retain and apply the strategies they learned during the intervention. There is a great deal of variability when it comes to

maintenance. In a recent systematic review, it was observed that most studies do report some maintenance but that this varied at the participant level (Wauters et al., 2023).

Overall, our findings are partially consistent with previous findings (Henry et al., 2018) wherein the authors reported significant improvement and maintenance of the production of correct, intelligible scripted words in individuals with nfvPPA following VISTA. In addition, our study extends previous findings by documenting, for the first time, the improvements in VISTA in lvPPA and svPPA while simultaneously documenting improvement across a more diverse set of outcome measures than previously reported, including keywords, use of synonyms, and global quality. With respect to intervention dosage, the findings reported by Henry et al. were a result of sessions of 45 minutes to one hour (twice per week) with four scripts trained over a four–six-week interval (Henry et al., 2018). In comparison, in the current study, a single script was trained in sessions of 30 minutes (twice per week) over an eight-week interval. As such, participants in the current study received a greater amount of training for a single script relative to Henry et al., although the duration of the individual intervention sessions was shorter. Taken together, this suggests that both shorter sessions and, potentially, a shorter duration of training could be feasible for a single script. Future studies investigating the effects of dosage on script training across PPA variants are needed.

In the current study, the results of a post-intervention Perceived Satisfaction/Acceptability Questionnaire, confirmed high satisfaction/acceptability as well as positive qualitative effects related to participants' overall experiences with therapy in terms of emotional and communication benefits. The benefits of script training include increased confidence in communication across the three variants of PPA. Whereas the majority of participants endorsed that they perceived benefits of intervention, two participants with svPPA reported neither agreeing nor disagreeing when asked if the therapy helped them emotionally. Because individuals with svPPA may present with some degree of difficulty with emotion

recognition and anosognosia (Savage et al., 2015; Valotassiou et al., 2022), it may be the case that these individuals were less aware of how the intervention was beneficial because of a lack of awareness about their communication impairments.

We also note that in the current study, no significant differences were observed based on intervention modality (in-person vs. teletherapy) with respect to intervention or generalization effects. These results are consistent with those of a previous study (Dial et al., 2019) reporting equivalent improvements in VISTA outcomes across intervention modalities. As such, teletherapy is feasible and can result in equivalent improvements in PPA compared with traditional in-person intervention, particularly for individuals in the mild to moderate stages of progression (Grasso et al., 2019; Rogalski et al., 2016; Schaffer et al., 2021).

4.2. Generalization effects

Generalization effects were also observed in the global quality ratings from pre-to post-intervention for the untrained, unscripted topic. Maintenance of this generalization effect was also observed from post-intervention to each follow-up. These findings partially align with those of Henry et al. , wherein the untrained scripts remained relatively stable during the follow-up period, although their outcome measures were more granular (e.g., overall intelligibility) relative to our study, wherein we analyzed ratings of the overall quality of the script production (e.g., intelligibility, fluency, vocabulary, and overall script coherence) (Henry et al., 2018).

With respect to our secondary outcome measures, a significant generalization effect was also observed on the naming task from the Cambridge Semantic Battery from pre- to post-intervention; however, this effect was not maintained when comparing the post-intervention performance to each follow-up. Naming improvements following script training have been reported in stroke-induced aphasia (Bilda, 2011; Grasso et al., 2019). Because individuals attempt to recall scripted words when producing scripts, an element of word selection and retrieval is foundational to script training and scripted content. The generalized improvement

documented in the naming task from the Cambridge Semantic Battery may therefore reflect enhanced word retrieval and selection beyond scripted words. Finally, with respect to MMSE, no non-significant changes were observed across time, indicating stability in general cognition during the duration of the study.

4.3. Effects and Relevance of Script Training by PPA Variant

We were particularly interested in investigating the effects of script training on these three variants of PPA. An analysis of individual responses to script training intervention revealed that significant and numerical improvements in script production and keywords were largely observed in individuals with lvPPA and nfvPPA. Additionally, small numerical improvements in synonym production were observed most consistently in lvPPA. The most robust and consistent improvements were observed in lv and nfvPPA, with less robust and less consistent improvements observed in individuals with svPPA.

Given the aforementioned pattern of results, we hypothesized that individuals with each of the PPA variants may respond differently to script training for different underlying reasons. In the case of nfvPPA, we observed that the benefits of the intervention were primarily in script and keyword production. We attribute these effects to the influence of script training on automatizing speech production, including the production of keywords (i.e., content words). Greater improvement in keywords reflects better production of content words relative to function words, with the latter being particularly challenging for individuals presenting with agrammatism and for those with motor speech impairment who may produce the most meaningful words to convey the intended message (Hoffman et al., 2017; Mesulam et al., 2014). We also note that although all participants with nfvPPA presented with clinical features of AOS, all individuals with nfvPPA demonstrated a robust response to the intervention.

In lvPPA, the enhanced script and keyword production is likely a result of lowering the demands associated with word retrieval when producing connected-speech. The improvement in synonym production reflects the use of alternative words with similar meanings, indicating that reliance on relatively spared semantic knowledge when impaired phonological processing diminishes the ability to recall a specific scripted word (Montembeault et al., 2018). Lastly, individuals with svPPA showed ceiling effects in fluency and script production, which limited our ability to capture the intervention effects. Nevertheless, individuals with svPPA demonstrated small numerical improvements in keywords and synonym production. This pattern reflects relatively spared fluency and phonology and variable improvements in the specificity and meaningfulness of their productions. Overall, our results indicate that individuals with different variants of PPA may benefit from script training in different ways, and that the effects differ on the basis of relatively spared versus impaired linguistic domains.

The effects of naming intervention in lvPPA and svPPA have been reported more frequently in the literature (Pagnoni et al., 2021). Many of these interventions focus on training single words, which may limit the ecological validity of these approaches, although some approaches train strategies as a means to promote generalization (Beales et al., 2016; Henry et al., 2019; Lavoie et al., 2020). In contrast to naming intervention, script training is a more ecologically valid approach when tailored to the individual, as it focuses on conveying personally relevant information that individuals are interested in communicating about *in conversation* (Hubbard et al., 2020). The philosophy of person-centeredness is considered integral to building a relationship with intervention participants and understanding their needs (Volkmer et al., 2022). Personalization of script content, as in the current study, is considered critical for engagement in learning (Cherney et al., 2015). Although this approach might be time-consuming for clinicians when creating intervention materials, personalization is a crucial component in providing relevant and motivating intervention programs for individuals living with PPA.

4.4. Strengths and Limitations of the Current Study

This study had several strengths and limitations. The strengths of the current study include reporting the effects of script training across PPA variants for the first time, as well as reporting preliminary patterns of intervention response by PPA variants. In addition, we investigated the effects of script training in the unique sociocultural context of Catalonia, with Spanish-Catalan bilingual participants (Grasso et al., 2023). Providing participants with the autonomy to select their language of intervention (which was always their dominant language), offers several benefits including a focus on the most functional language which increases engagement and motivation. Receiving intervention in your dominant language ensures that the intervention is more culturally relevant as the use of cultural expressions, and communication styles are incorporated into the intervention. Given that both in-person intervention and teletherapy offers positive benefits, the broader use of teletherapy in PPA will allow greater access to specialized services, with greater likelihood of offering intervention in participants' language of preference.

Demonstrating the acceptability and feasibility of modified VISTA in a novel context indicates that this approach may be acceptable across distinct sociocultural contexts, as the development of materials is personalized for each participant, including their cultural and linguistic backgrounds.

With respect to limitations, our sample was relatively small, particularly when examining patterns by the PPA variant. Given our small sample of participants with nfvPPA, we also were unable to discern how severity of motor speech impairment might impact response to intervention, though all participants with nfvPPA showed a robust response to intervention in this study. Second, consistent with previous research investigating the effects of script training on PPA, we only included individuals in the mild-to-moderate stages of disease progression. Therefore, the effects of script training in more advanced participants cannot be discerned from

the current study. In addition, our untrained topic was not scripted; therefore, we could not ensure that the untrained content was matched for linguistic complexity with that of the trained script. We also did not have an objective approach for quantifying the amount of home practice individuals engaged in; therefore, the intensity of home practice may have differed across participants and ultimately influenced some of the effects reported herein. Lastly, although we assessed acceptability using an in-house developed measure, with the goal of using aphasia-friendly language, the use of additional questionnaires may have more robustly captured acceptability and future studies may consider using more than one tool to more rigorously assess acceptability.

4.5. Conclusion

Our study showed that the modified version of VISTA used in this study is a feasible and acceptable intervention for individuals with the three PPA variants, with the most robust and consistent improvements observed in lv and nfvPPA. We observed positive effects across a variety of outcome measures, including script production, keywords, use of synonyms, global quality, and improvement in a measure of confrontation naming. Participants also reported subjective improvements in communication and emotionality. Larger studies are needed to further elucidate the unique effects of script training on each PPA variant. Efforts to support the wider-scale implementation of script training in PPA are needed, as this approach represents a functional, person-centered intervention option that supports individuals' abilities to continue communicating about topics of personal relevance in their everyday lives.

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Data Availability Statement

The datasets generated in the current study are available from the corresponding author upon reasonable request.

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Table 1

Demographic and cognition, speech and language scores at baseline

PARTICIPANT ID	P01	P02	P03	P04	P05	P06	P07	P08	P09	P10	P11	P12	P13
Demographics													
Age	74	61	74	74	66	67	72	66	77	66	81	67	74
Sex	Female	Female	Female	Male	Female	Female	Female	Female	Female	Female	Male	Female	Male
Education (yr)	6	12	16	16	6	12	12	12	16	12	16	16	6
PPA variant	svPPA	svPPA	svPPA	svPPA	svPPA	lvPPA	lvPPA	lvPPA	lvPPA	lvPPA	nfvPPA	nfvPPA	nfvPPA
Aphasia Severity	Moderate	Mild	Moderate	Mild	Moderate	Moderate	Moderate	Mild	Moderate	Mild	Mild	Moderate	Moderate
Intervention modality	P	T	P	P	T	P	T	T	P	P	T	T	T
Language	Catalan	Spanish	Catalan	Catalan	Spanish	Catalan	Spanish	Catalan	Spanish	Catalan	Spanish	Catalan	Spanish
Cognitive-Linguistic Scores													
MMSE/30	20	27	19	25	21	15	18	30	21	28	30	27	28
nCSB/64	22	40	9	41	18	41	42	60	56	58	57	62	62
Digit span/26	8	14	10	14	14	5	8	11	8	12	13	8	15
BSBA (seconds)	107	172	139	158	164	244	185	115	313	156	268	390	369

Abbreviations: lvPPA: logopenic variant of primary progressive aphasia; nfvPPA: non-fluent variant of primary progressive aphasia; svPPA: semantic variant of the PPA; P: in-person; T: teletherapy; MMSE: Mini-Mental State Examination; nCSB: Naming task from the Cambridge Semantic Battery; BSBA: Barcelona Scale for Buccophonatory Apraxia (greater duration=worse performance)

Table 2

Original VISTA Treatment Steps

1. Recognize script sentences from foils
1. Put script sentences in order
2. Read the entire script aloud
3. Produce script sentence in response to questions
4. Produce the entire script from memory
5. Respond to questions with scriped sentences (not in scripted order)
6. Conversation with naïve communication partner

Modified VISTA Treatment Steps Used in Current Study

1. Read aloud each scripted sentence in response to question posed by clinician, in order of scripted sentences (Step 4 in original VISTA)
2. Read aloud each scripted sentence in response to question posed by clinician, out of scripted order (Step 6 in original VISTA)
3. Read the entire script aloud (Step 3 in original VISTA)
4. Produce each scripted sentence in response to question posed by clinician, in order of scripted sentences (Step 4 in original VISTA)
5. Produce the entire script from memory (Step 5 in original VISTA)
6. Produce each scripted sentence in response to question posed by clinician, out of scripted order (Step 6 in original VISTA)
7. Produce each scripted sentence in response to question posed by peer with PPA, out of scripted order (Closest to Step 7 in original VISTA)

Table 3

Individual participant difference scores for trained content from baseline/pre-intervention relative to each follow-up

		SCRIPT PRODUCTION			**KEYWORD PRODUCTION**			**SYNONYM PRODUCTION**		
Participant	Variant	T0-T1	T0-T1	T0-T3	T0-T1	T0-T1	T0-T3	T0-T1	T0-T1	T0-T3
P01	svPPA	-1%	0%	0%	6%	6%	-12%	13%^	13%^	0%
P02	svPPA	-1%	0%	0%	26%	0%	0%	0%	0%	0%
P03	svPPA	-1%	-1%	-1%	0%	9%	0%	0%	0%	0%
P04	svPPA	0%	0%	0%	0%	12%	16%	8%	4%	0%
P05	svPPA	3%	3%	3%	79%*	68%*	16%	0%	0%	0%
P06	lvPPA	22%*	17%*	12%	34%*	22%	17%	11%	5%	11%
P07	lvPPA	-8%	-5%	0%	32%*	27%*	14%	5%	9%	5%
P08	lvPPA	5%	-8%	5%	48%*	51%*	45%*	4%	7%	7%
P09	lvPPA	4%	11%	5%	80%*	80%*	70%*	5%	5%	15%*
P10	lvPPA	11%	5%	14%*	44%*	52%*	32%*	4%	0%	0%
P11	nfvPPA	1%	-21%	-15%	55%*	42%*	38%*	13%*	13%*	8%
P12	nfvPPA	4%	1%	0%	63%*	47%*	47%*	0%	-5%	0%
P13	nfvPPA	11%	7%	0%	66%*	71%*	71%*	0%	5%	5%
MEAN		4%	1%	2%	41%	37%	27%	5%	4%	4%
MIN		-8%	-21%	-15%	0%	0%	-12%	0%	-5%	0%
MAX		22%	17%	14%	80%	80%	71%	13%	13%	15%

*Note. Significance testing conducted via simulation analyses at the single participant level. *Denotes significant change at the single participant level; ^ Denotes marginal change at the single participant level. T0= Baseline/pre-intervention; T1= post-intervention; T2= 3-months post-intervention and T3= 6-months post-intervention.

FIGURES

Figure 1. Accuracy of Script Production by Intervention Modality Across Time.

*Note: Results of permutation tests comparing effects between participants who received intervention via in-person vs. teletherapy at each time point. T0 = baseline; T1 = post-intervention; T2 = 1-month post-intervention; T3 = 3 monthspost-intervention. t = statistic test

Figure 2. Primary Outcome Measures for Trained and Untrained Content Across Time

*Note. Means, standard errors, and individual-level performances are depicted for each outcome measure. Significance was determined via one-tailed permutation tests for trained content from the baseline to each subsequent time point. Significance was determined using two-tailed permutation tests from post-intervention to each subsequent time point. T0 = baseline; T1 = post-intervention; T2 = 1-month post-intervention; T3 = 3 months post-intervention. * = $p \leq .05$; ** = $p \leq .01$, *** $p \leq .001$.

Figure 3. Performance on Secondary Outcome Measures Over Time.

*Note. Means, standard errors, and individual-level performance were depicted for each secondary outcome measure. Significance was determined via two-tailed permutation tests for the Cambridge Naming Test and MMSE from baseline to each subsequent time point, and from post-intervention to each subsequent time point. T0 = baseline; T1 = post-intervention; T2 = 1-month post-intervention; T3 = 3 months post-intervention. * = $p \leq .05$; ** = $p \leq .01$, *** $p \leq .001$.

Figure 4. Average Participant Ratings in Response to Post-Intervention Perceived Satisfaction/Acceptability Questionnaire

*Note:1 = strongly disagree; 2 = disagree; 3 = neither agree nor disagree; 4 = agree; and 5 = strongly agree.

SUPPLEMENTAL MATERIAL

Script trained example

Perceived Satisfaction/Acceptability Questionnaire