Aphasiology

ISSN: (Print) (Online) Journal homepage: https://www.tandfonline.com/loi/paph20

Preserving lexical retrieval skills across languages in a bilingual person with logopenic primary progressive aphasia

Aviva Lerman, Dorit Mais, Yael Nissani & Taryn Malcolm

To cite this article: Aviva Lerman, Dorit Mais, Yael Nissani & Taryn Malcolm (2022): Preserving lexical retrieval skills across languages in a bilingual person with logopenic primary progressive aphasia, Aphasiology, DOI: 10.1080/02687038.2021.2020717

To link to this article: https://doi.org/10.1080/02687038.2021.2020717

APHASIOLOGY
https://doi.org/10.1080/02687038.2021.2020717

Preserving lexical retrieval skills across languages in a bilingual person with logopenic primary progressive aphasia

Aviva Lerman[a], Dorit Mais[a], Yael Nissani[a] and Taryn Malcolm[b]

[a]Program in Communication Disorders, Hadassah Academic College, Jerusalem, Israel; [b]Department of Communication Disorders, Mercy College, New York, NY, USA

ABSTRACT

Background: The treatment of lexical retrieval in monolingual people with the logopenic variant of primary progressive aphasia (lvPPA) has been observed to preserve or improve skills to varying degrees. There is a paucity of treatment literature for multilingual people with PPA (across all types), although based on the stroke-induced aphasia literature we would expect treatment to be effective in the treated language and potentially the untreated language too.

Aims: We investigated the effects of a verb-based semantic treatment administered in a later-acquired language to an English-Hebrew speaker with lvPPA on her lexical retrieval skills in different language tasks in both the treated and untreated languages.

Methods & Procedures: Language skills across different tasks were assessed pre- and post-treatment, with Verb Network Strengthening Treatment (VNeST) provided in Hebrew. We evaluated whether decline continued for lexical retrieval (as observed in the years leading up to the study), and in which language(s).

Outcomes & Results: We observed that lexical retrieval skills in both languages did not decline for word production, sentence production, and written narratives, but did continue to decline during oral narrative production.

Conclusions: Our results indicate that VNeST may be an effective prophylactic treatment for the preservation of lexical retrieval skills in both a treated and untreated language of multilingual people with the logopenic variant of PPA.

ARTICLE HISTORY
Received 6 April 2021
Accepted 16 December 2021

KEYWORDS
Logopenic variant primary progressive aphasia; multilingual; Verb Network Strengthening Treatment; lexical retrieval; cross-language generalisation

Introduction

Logopenic variant of primary progressive aphasia

Primary progressive aphasia (PPA) is a degenerative neurological condition where language abilities slowly and gradually decline. There are three main subtypes of PPA: the logopenic variant (lvPPA), the semantic variant (svPPA) and the non-fluent variant (nfvPPA). The logopenic variant of PPA has linguistic deficits often attributed to damage

CONTACT Aviva Lerman avivale@edu.hac.ac.il Program in Communication Disorders,Hadassah Academic College, Jerusalem, Israel
Supplemental data for this article can be accessed here.

initially occurring in the phonological network. Thus, the most common language impairments in the early stages of lvPPA are word-finding difficulties, poor repetition skills (especially for sentences relative to single-words), and dysfluency in oral narratives (Gorno-Tempini et al., 2011; Mesulam, 2007). Furthermore, phonemic paraphasias and literal paragraphias are common across different language tasks, while comprehension and grammatical skills are expected to be relatively spared, at least initially (Gorno-Tempini et al., 2011; Mesulam, 2007).

The general underlying etiology of lvPPA is attributed to atrophy or hypoperfusion in the left posterior temporoparietal region of the brain (Gorno-Tempini et al., 2011; Mesulam, 2007). However, patients diagnosed with lvPPA show clinical heterogeneity relative to specific etiologies, as observed by metabolic patterns on positron emission tomography (PET) scans (Krishnan et al., 2016). Krishnan and colleagues found three different etiologies in a group of amyloid-positive lvPPA patients, that they labelled according to where predominant hypometabolism was observed: temporal, parietal, and temporoparietal subgroups.

All three subgroups revealed language decline patterns appropriate to an lvPPA diagnosis, but the parietal subgroup (and to a lesser extent the temporoparietal subgroup) also showed salient non-linguistic cognitive deficits such as verbal working memory impairment, executive function decline, and reduced mental flexibility, with the parietal group exhibiting behavioural dysfunction too. The temporal lvPPA subgroup was least associated with non-linguistic cognitive deficits (Krishnan et al., 2016). Thus, although the phonological network may be initially impaired in people with lvPPA, as the disease progresses and more linguistic and non-linguistic networks degenerate, word finding difficulties become more pronounced and other relatively spared language skills start to decline (Henry et al., 2019; Krishnan et al., 2016; Win et al., 2017).

Treating primary progressive aphasia

Treating PPA is challenging due to its degenerative nature and late diagnosis relative to symptom onset. Treatment is considered beneficial not only when improvement occurs (i.e., remediation) but also when decline does not occur (i.e., prophylaxis; e.g., Henry et al., 2019; Meyer et al., 2015, 2018). Generally, language intervention may be beneficial for about 6- to 12-months following treatment (Tippett et al., 2015). However, generalisation is affected by PPA subtype. For example, better generalisation for lvPPA and nfvPPA has been observed compared with svPPA (Cadório et al., 2017). Furthermore, factors such as continued practice, length of treatment and frequency of sessions will affect treatment outcomes (e.g., Cadório et al., 2017).

Lexical retrieval impairment refers to word retrieval difficulties, which are a prominent symptom of aphasia and have been noted as a key feature of lvPPA (Edmonds et al., 2009; Laine & Martin, 2006). Impairments in lexical retrieval can impact language for single-words, sentences, and discourse (e.g., Edmonds, 2016). In stroke-induced aphasia, verb- or noun-based semantic treatment is often administered to improve lexical retrieval, regularly resulting in generalisation to untrained stimuli and/or untrained contexts (e.g., Edmonds, 2016; Kiran & Bassetto, 2008; Nickels, 2002; Raymer & Ellsworth, 2002; Wambaugh et al., 2002). In lvPPA, several treatment studies have investigated lexical retrieval in monolingual individuals, utilizing a variety of interventions (e.g., semantic, phonological, and/or orthographic treatments). These interventions often focus on noun retrieval (e.g., Beeson et al., 2011; Henry et al., 2019; Newhart et al., 2009), even when salient verb deficits are observed (e.g., Newhart et al., 2009), although some have focused on lexical retrieval of different word classes, such as verbs and adjectives together with nouns (e.g., Beales et al., 2016). Results indicate either preserved lexical retrieval skills over the course of a treatment block (e.g., Meyer et al., 2016; 2018), or improvement retrieving treated stimuli, with some generalisation to untreated stimuli in a few cases (e.g., Beales et al., 2016; Beeson et al., 2011; Henry et al., 2013; 2019; Newhart et al., 2009; Rapp & Glucroft, 2009).

Meyer et al. (2020) suggest that individuals with PPA who have a salient action naming impairment may benefit from a verb-based semantic treatment, such as Verb Network Strengthening Treatment (VNeST), which requires participants to retrieve pairs of agents (subjects) and patients (objects) with a given verb to produce relevant SVO sentences. VNeST was developed with the aim of strengthening the conceptual connections between verbs and their thematic roles in order to increase lexical retrieval of content words in sentences and connected speech in people with aphasia (Edmonds, 2016; Edmonds & Babb, 2011; Edmonds et al., 2009). The strengthening of these conceptual connections was hypothesised to result from Hebbian strengthening of neurological connections occurring from repeated activation of different verbs together with a variety of relevant thematic roles. Thus, VNeST should slow the decline of lexical retrieval in lvPPA due to either remediation of declining lexico-semantic connections, or compensation of declining lexico-phonological connections by allowing for more efficient access to the semantic network. Indeed, Gorno-Tempini et al. (2008) observed that people with lvPPA rely more on a semantic route than a phonological route when repeating sentences, based on their patterns of errors, supporting this hypothesis of lexico-phonological compensation.

Primary progressive aphasia and multilingualism

Two review papers examined patterns of language decline in multilingual people with PPA (Costa et al., 2019; Malcolm et al., 2019) and both papers concluded that language decline occurs in both the first-acquired language (L1) and a later-acquired language (L2) but not necessarily in a parallel manner. Furthermore, the first symptom of language decline was most commonly word retrieval impairment in L2 (Costa et al., 2019). Five case-studies measuring language decline longitudinally, but without any intervention, were included in the review papers (Druks & Weekes, 2013; Filley et al., 2006; Larner, 2012; Liu et al., 2012; Machado et al., 2010). In all five cases, L2 declined before or in parallel with L1, but as the disease progressed, both L1 and L2 were impaired in a similar manner.

Overall, these patterns of decline are independent of age of acquisition, manner of acquisition, or daily language use (Malcolm et al., 2019) but PPA subtype, task type, and language dominance may influence impairment patterns across languages (Costa et al., 2019). For example, similar impairments for naming difficulties were observed for L1 and L2 in lvPPA and svPPA but not for nfvPPA where L2 was always observed to be more impaired than L1 regardless of dominance (Costa et al., 2019). Additionally, language repetition skills were observed to be similar across L1 and L2 in all subtypes, but tasks related to semantic knowledge were more impaired in L2 than L1 for svPPA and nfvPPA subtypes only (Costa et al., 2019).

When exploring language decline in multilingual people with lvPPA, we must also consider the underlying neural substrates. In people with PPA, as neurons degenerate in language areas of the brain, spared neurons and white matter pathways can partially reorganize to retain some language function, particularly in the early stages of the disease (e.g., Malcolm et al., 2019; Mesulam et al., 2014). Thus, although language continues to decline over time due to cortical atrophy, this reorganisation contributes to the gradual nature of the decline that is so typical of PPA. Why language decline may be differential or parallel across L1 and L2 in multilingual people with PPA can therefore be considered in terms of shared or separated neural substrates subserving each language.

Cortical representations of different languages have been shown to largely overlap in multilingual people (e.g., Fabbro, 1999; Halsband, 2006; Marian et al., 2003; Van de Putte et al., 2018; Yang et al., 2017), especially when they are highly proficient (e.g., Videsott et al., 2010). However, within overlapping cortical areas, distinct neural circuits are hypothesised to independently subserve different languages (e.g., García-Pentón et al., 2014; Halsband, 2006; Wong et al., 2016). Thus, the extent of overlap within neural substrates and circuits subserving each language should determine the degree that neural atrophy and/or any subsequent neural reorganisation will affect the different languages in parallel or differentially across language tasks.

Based on Hebbian theory, neural networks engaged more often for a certain linguistic process would retain better plasticity and subsequent synaptic strengthening than those engaged less frequently (e.g., Berthier & Pulvermüller, 2011; Fox & Stryker, 2017). Thus, generally, neural circuits subserving a later-acquired, less-dominant language would be expected to break down at a faster rate than those subserving an early-acquired, more dominant language, either in cortical areas with minimal overlap across languages, or in distinct neural circuits within largely overlapping cortical areas. However, as the disease progresses and reorganisation of spared neurons and white matter pathways becomes less viable, both languages should be impaired in a similar way due to more widespread cortical atrophy. Patterns of decline observed in multilingual people with PPA support these theories of parallel vs. differential language decline due to the effect of cortical degeneration, because decline is often parallel across languages, certainly in the later stages. Furthermore, if one language does decline faster than another, the better-spared language will be the L1 and/or dominant language (Costa et al., 2019; Malcolm et al., 2019).

Regions and language networks associated with semantic and phonological processing have been observed to overlap more than those associated with lexical processing in different languages (e.g., Marian et al., 2003; Van de Putte et al., 2018; Yang et al., 2017), a finding that supports theoretical models of bilingualism that hypothesise shared conceptual representations together with partially separate lexicons (e.g., Kroll & Tokowicz, 2005; Kroll et al., 2010; Nadeau, 2019; Paradis, 1993; Van Hell & De Groot, 1998). Furthermore, we might expect that connections in neural networks involving lexical knowledge would be reinforced only within language, but that connections in neural networks involving semantic knowledge would be reinforced across all languages. Thus, we might expect that tasks relying on semantic processing would decline in parallel across languages, but those related to lexical processing would be more susceptible to differential decline. Currently, empirical data does not fully support this distinction across PPA subtypes, but there is partial support from data within the lvPPA subtype (Costa et al., 2019).

Treating primary progressive aphasia in multilingual people

Based on Hebbian theory discussed above, we would also expect (and observe in monolingual people with PPA) that specific language processes targeted in therapy would be better preserved than those not targeted (Berthier & Pulvermüller, 2011; Fox & Stryker, 2017). For multilingual people with PPA, we would expect different outcomes depending on the interaction of PPA subtype (and underlying neuronal damage), relative language proficiencies, and treatment type.

Within a framework of shared neural substrates subserving semantic knowledge, we might expect semantic treatment in either language to prevent or slow the decline of lexical retrieval for both languages because lexico-semantic connections will potentially be strengthened, similar to within- and cross-language generalisation observed post-stroke after semantic treatment (e.g., Goral & Lerman, 2020; Kiran et al., 2013; Lerman et al., 2018, in press). This would be especially probable in lvPPA in later stages of the disease if atrophy spreads through the temporal lobe towards the temporal pole, directly affecting semantic networks. However, even if damage is confined to the phonological network, semantic treatment that involves repeated naming practice, such as VNeST, could still be effective in both languages due to potentially stronger lexico-semantic connections reducing the impact of a declining phonological network, especially in highly proficient multilingual people.

Conversely, within a framework of distinct neural circuits for each language, we might expect semantic treatment in one language to prevent or slow decline in the treated language only. For example, if the primary impairment is in the phonological network, with weak lexico-phonological connections, difficulties would likely be language-specific. Semantic treatment that strengthens lexico-semantic connections may not be effective enough to compensate for these lexico-phonological difficulties in the untreated language, particularly once these impairments degenerate past a certain point. This may be particularly relevant if semantic treatment is administered in L1 – treatment effects may be unlikely to generalise to a less-dominant L2 if lexico-semantic connections between the L2 and the semantic network are degenerating faster than between the more-dominant L1 and the semantic network, as expected in multilingual people with PPA (Costa et al., 2019; Malcolm et al., 2019). Alternatively, semantic treatment in L2 that strengthens lexico-semantic connections may still generalise to the relatively spared L1 and result in better L1 lexical retrieval after treatment. Finally, phonologically focused treatment may be effective in the treated language only. Treatment effects would be less likely to generalise to an untreated language, due to the language-specific nature of lexico-phonological connections.

To date, only one treatment study has been published on a bilingual participant with PPA (Meyer et al., 2015). Meyer and colleagues investigated treatment efficacy for anomia in a treated language, and generalisation to an untreated language, in a 69-year-old Norwegian-English bilingual participant with lvPPA. Pre-morbidly, she was highly proficient in both languages, with Norwegian her native language, and English acquired from age 7-years, and becoming the language of her environment for over 40 years. The decline of her two languages was mostly parallel at baseline testing across different language tasks. Both orthographic and phonological treatment were administered in English only over a 1-year period, with eight treatment sessions in the first month, followed by one session a month for 11 months, with three home practices a week (Meyer et al., 2015).

Meyer and colleagues found that orthographic treatment resulted in greater English written naming accuracy of trained words (which was not maintained after treatment ended) with generalisation to Norwegian as observed by relatively more accurate oral naming and naming to definition of the translation of words trained orthographically than words trained phonologically or untrained (Meyer et al., 2015). However, overall naming in Norwegian declined from baseline to post-treatment, so the contribution of treatment in English to Norwegian is inconclusive. Furthermore, while phonological treatment resulted in greater English oral naming accuracy of trained words, no effect on these words was observed for naming accuracy in Norwegian (Meyer et al., 2015). The authors concluded that because generalisation to the untreated language was observed after orthographic treatment but not after phonological treatment, orthographic treatment probably strengthened the shared semantic network more than phonological treatment strengthened it (Meyer et al., 2015). However, because both treatment types were provided simultaneously, it is unclear how each may have influenced the other (i.e., cumulative effects vs. separate effects), and thus how each type of treatment affected retrieval ability relative to the underlying impairment.

The current study

We investigated the effect of VNeST on lexical retrieval of a participant with lvPPA (whose atrophy had spread from left temporoparietal areas to the anterior temporal pole) across different language tasks at the word, sentence, and discourse levels. We chose VNeST as our preferred semantic treatment for two reasons. First, because at baseline testing our participant's action naming (AN) was significantly worse than her object naming (ON) (see, Table 4), thus VNeST may be considered a potential treatment option for this participant (Meyer et al., 2020). Second, because VNeST has been observed to result in generalisation across languages for stroke-induced aphasia in multilingual people (Lerman et al., in press), providing support for its effects on strengthening the shared semantic network.

We asked the following research questions:

(1) In a bilingual individual with lvPPA, does providing VNeST in the post-morbidly more-impaired L2 interrupt the deterioration of lexical retrieval in that language?
(2) Do any treatment effects generalise to the untreated post-morbidly less-impaired L1?

We hypothesised that VNeST would interrupt the deterioration of lexical retrieval in single-word, sentence, and discourse tasks in the treated language due to strengthening the semantic network (Edmonds, 2016; Edmonds & Babb, 2011; Edmonds et al., 2009). A strengthened semantic network should allow continuing access to the lexicon, potentially strengthening lexico-semantic connections and improving lexical retrieval (Berthier & Pulvermüller, 2011; Fox & Stryker, 2017; Meyer et al., 2020).

We also hypothesised that the untreated language should also benefit from VNeST in similar lexical retrieval tasks within a framework of shared neural substrates subserving semantic knowledge (Paradis, 1993). Furthermore, due to the spreading atrophy from left temporoparietal areas to the anterior temporal pole, we do not expect degeneration only across lexico-phonological connections, rather we expect degeneration throughout the shared semantic network and its lexico-semantic connections too, and therefore impairments and potential treatment effects should generalise across languages, at least partially. Finally, by treating and strengthening the semantic network via the more-impaired L2, the effects of treatment should generalise to the less-impaired L1.

Methodology

Ethics approval was obtained from the Hadassah Academic College Ethics Committee prior to recruitment. Written informed consent was obtained by providing a combination of clear verbal and written information about the study.

Participant

The participant was a 70-year-old female with over 20 years of formal education, who used both English and Hebrew daily in both her home environment and her academic life. English was her first-acquired language (L1), from birth. Hebrew was a later-acquired language, beginning from age 8 years in school and reaching high proficiency – including literacy – at around age 20 years, after moving to a Hebrew-speaking environment. The participant reported pre-morbid high proficiency in both English and Hebrew in all modalities, although English remained her dominant language across the lifespan. She also reported acquiring French as a child (before acquiring Hebrew) but not having used French in her daily life for over 30 years. French was not assessed or treated during this study. Thus, we refer to Hebrew as her L2 in this paper.

Ten years prior to taking part in this study, the participant noticed minimal difficulties with word retrieval, a common first symptom in PPA (Westbury & Bub, 1997), specifically in Hebrew (her L2). In 2010, she was examined by a neurologist who found language and cognitive skills to be within normal limits, with no abnormal findings during brain imaging. Her language skills continued to deteriorate, again specifically in Hebrew, and in 2014, a PPA diagnosis was raised as a possible explanation. A PET/CT scan from 2015 showed hypoperfusion in left temporoparietal and anterior temporal lobe regions, with possible left frontal lobe hypoperfusion as well. However, she was only officially diagnosed with lvPPA in 2018.

This diagnosis was confirmed by the authors using reports on speech and language assessment and treatment from her medical file (she provided consent for the researchers to access it) as well as by her relative linguistic and non-linguistic abilities at the time of

Table 1. Self-reported language skills in English and Hebrew before the onset of PPA and at the time of the study (10 years after the onset of PPA symptoms).

	Speech		Comprehension		Reading	
	Pre-morbid	Post-morbid	Pre-morbid	Post-morbid	Pre-morbid	Post-morbid
English (score out of 10)[a]	10	7	10	8	10	9
Hebrew (score out of 10)[a]	9	3	9.5	6	9.5	7

[a]10 = native-like proficiency, 0 = no ability

participation in our study. First, the participant reported that during the 10 years prior to our study, her principal deterioration was for language, and those deteriorating language skills negatively impact daily living activities. This report, together with her non-linguistic cognitive skills measuring within normal limits for her age at the time of the study (see Supplementary Table 1), supports the diagnosis based on PPA diagnostic criteria (Gorno-Tempini et al., 2011; Mesulam, 2001). More specifically, the participant reported that her L2 Hebrew language skills deteriorated faster than her L1 English language skills, as expected in multilingual people with PPA (Costa et al., 2019; Malcolm et al., 2019). See Table 1 for self-reported language skills. She also reported that while daily production of Hebrew and English was equal before deterioration began (50% of each day), at the time of the study daily production of English was higher than Hebrew (90% English, 10% Hebrew).

Second, the participant was initially assessed by a speech and language therapist (SLT) in Hebrew in 2014 after a referral by her neurologist, and received treatment once a week, in Hebrew, between 2015 and 2019 (until 1-month prior to the study). Based on her Western Aphasia Battery-Revised (WAB-R) scores in both languages it was observed that over the five years prior to the study her Hebrew indeed deteriorated more and at a faster rate than her English (see from August 2015 to July 2019 in Figure 1), with her overall Aphasia Quotient (AQ) at the beginning of the current study (July 2019) indicating moderate aphasia in Hebrew (60.2/100) and mild aphasia in English (91.4/100). Deterioration was not equal across all subtests (spontaneous speech, auditory comprehension, repetition, and naming and word-finding) rather, as expected for the lvPPA subtype (Gorno-Tempini et al., 2011; Mesulam, 2001), the most salient deterioration was observed in both languages for repetition, followed by spontaneous speech and naming

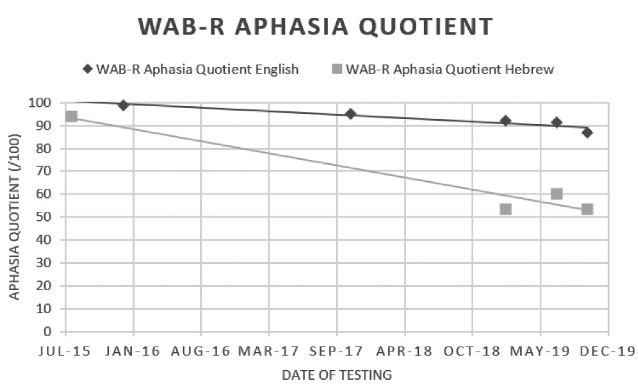

Figure 1. WAB-R Aphasia Quotients in Hebrew and in English between 2015–2019.

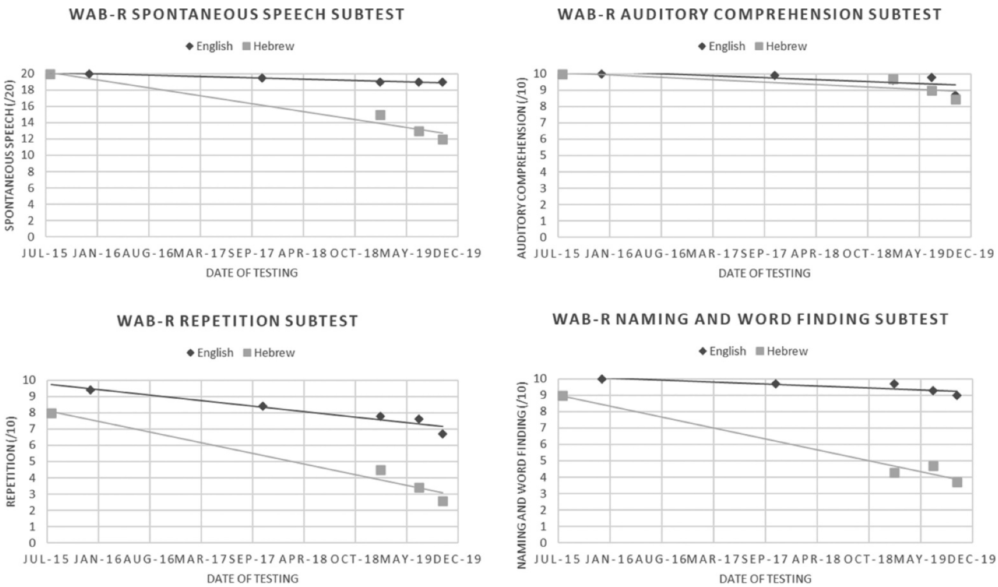

Figure 2. WAB-R scores per subtest in Hebrew and in English between 2015–2019.

and word-finding in Hebrew more than English. Furthermore, relatively preserved single-word auditory comprehension that is also expected in the lvPPA subtype (Gorno-Tempini et al., 2011) was observed for the participant whose auditory comprehension deteriorated minimally in Hebrew only (see from August 2015 to July 2019 in Figure 2).

Finally, we note that deterioration of language skills can continue for many years without deterioration in non-linguistic cognitive skills (Mesulam, 2001; Westbury & Bub, 1997). Thus, the slow deterioration of language over a period of 10 years that was observed in our participant is not unusual.

Procedure

This study takes a multiple-baseline case study approach with three phases: (1) pre-treatment baseline testing in both Hebrew and English, (2) Hebrew treatment block (VNeST), and (3) post-treatment testing of both Hebrew and English. Pre-treatment testing scores for each language provided a baseline for the participant to which post-treatment testing scores were compared.

Procedure: pre- and post-treatment assessments

English and Hebrew language skills were assessed at two time-points – pre-treatment (baseline – July 2019) and post-treatment (October 2019). At each time-point, language skills were assessed using the WAB-R in English (Kertesz, 2006) and in Hebrew (using a non-standardised adaptation of the WAB-R; Soroker, 1997), and a comprehensive aphasia battery that was developed specifically for English-Hebrew speakers (the REHAB – Revised English-Hebrew Aphasia Battery; Lerman & Goral, unpublished). The subtests in the REHAB are comparable across languages based on several linguistic and psycholinguistic factors such as frequency, argument structure, number of translation equivalents, cognate status, etc. In this study, we used production subtests from the

Table 2. Revised English-Hebrew Aphasia Battery subtests, measures and stimuli development (Lerman & Goral, unpublished).

Subtest	Measures	Task	Stimuli development
Object naming (n = 45)	Lexical retrieval accuracy of single-word nouns	Participants were asked to name pictures of objects	Partially based on a subset of pictures from the Multilingual Naming Test (Gollan et al., 2012); whole set of pictures piloted on two healthy speakers of English and two healthy speakers of Hebrew
Action naming (n = 45)	Lexical retrieval accuracy of single-word verbs	Participants were asked to name pictures of actions	Partially based on a subset of pictures from the Action Naming Test, which is a subset of the Verb and Sentence Test (Bastiaanse et al., 2002); whole set of pictures piloted on two healthy speakers of English and two healthy speakers of Hebrew
Sentence construction 1-argument (n = 12) 2-arguments (n = 30) 3-arguments (n = 12)	Lexical retrieval accuracy within sentences as measured by relevant SVO sentence production (= CUs)	Participants were asked to describe each picture using a single sentence	Partially based on a subset of pictures from the Object and Action Naming Battery (Druks & Masterson, 2000); whole set of pictures piloted on two healthy speakers of English and two healthy speakers of Hebrew
WH-questions (n = 16)	Functional language sentence production as measured by the accuracy of relevant, complete answers	Participants were asked to answer an everyday question, presented verbally, in one sentence	Based on a set of questions developed for testing English and Hebrew speakers with aphasia by Goral and Borodkin (unpublished)
Discourse – oral: Picture description (n = 6) 6-picture story sequence (n = 3) Personal narrative (n = 3) Procedural narrative (n = 3)	Lexical retrieval accuracy within utterances measured by calculating the percentage of relevant, SVO utterances out of all utterances (%CUs) Lexical diversity as measured by noun type-tokens ratios and verb type-tokens ratios	Participants were asked to describe pictures, tell stories based on 6-pictures, tell personal narratives, and tell procedural narratives	Developed and piloted on 12 healthy speakers: f = 7, m = 5; native speakers of English = 7, native speakers of Hebrew = 5; ages 57–76 years (mean = 68.3 years); 11–25 years of formal education (mean = 16.5); middle to high SES Included picture descriptions of the "Cookie Theft" from the Boston Diagnostic Aphasia Examination (Goodglass & Kaplan, 1983) and the "picnic" from the WAB-R (Kertesz, 2006), and also the 6-picture story sequence from the Bilingual Aphasia Test (Paradis, 2011)
Discourse – written Picture description (n = 6) 4-picture story sequence (n = 3) Personal narrative (n = 3) Procedural narrative (n = 3)	Lexical retrieval accuracy for written sentences measured by calculating the percentage of relevant, SVO sentences out of all sentences (%CSs) Lexical diversity as measured by noun type-tokens ratios and verb type-tokens ratios	Participants were asked to write descriptions of pictures, write stories based on 4-pictures, write personal narratives, and write procedural narratives	Developed and piloted on 12 healthy speakers: f = 7, m = 5; native speakers of English = 7, native speakers of Hebrew = 5; ages 57–76 years (mean = 68.3 years); 11–25 years of formal education (mean = 16.5); middle to high SES Included 4-picture story sequences from Goral & Borodkin (unpublished)

(*Continued*)

Table 2. (Continued).

Subtest	Measures	Task	Stimuli development
Non-word repetition (control) (n = 30)	Control task: accuracy measure of non-word repetition	Participants were asked to repeat non-words	The non-word repetition subtest from the Psycholinguistic Assessments of Language Processing Aphasia (PALPA) in English (Kay et al., 1996) and in Hebrew (Gil & Edelstein, 2001)

Note. CU = Complete Utterance – an utterance relevant to the stimulus that contained a subject and a verb (and an object, when necessary). CS = Complete Sentence – a written sentence relevant to the stimulus that contained a subject and a verb (and an object, when necessary). SES = socioeconomic status.

REHAB that included single-words, sentences, and discourse, as well as non-word repetition. See, Table 2 for a detailed summary of each subtest (what was measured and how we developed them).

The participant was assessed in both English and Hebrew at pre-treatment over five days and at post-treatment over four days. On each assessment day, at each time-point, one-third of the REHAB was administered in one or both languages. Each third was comparable to the others for each subtest per language, based on both the number and type of items per test and psycholinguistic factors for specific stimuli such as word length, frequency, etc. Thus, we conducted multiple pre- and post-treatment testing sessions while reducing task repetition, a methodology shown to be reasonable and necessary when assessing language deficits across many languages using long and potentially fatiguing testing batteries (Borodkin et al., 2020). The WAB-R was administered once in each language at pre-treatment and at post-treatment.

When more than one language was assessed on any given day, long breaks were taken between languages, and order of language testing was counterbalanced. Assessment was carried out by two SLT students who were trained on the assessment procedure. Each assessor tested only one language, but both were highly proficient in both English and Hebrew. Assessment sessions were videotaped and later transcribed by the same two SLT students who assessed the languages. Each student transcribed both languages independently, and then compared transcriptions. A small number of discrepancies were mediated by the P.I. (A.L.). Overall, accuracy of transcriptions was found to be high, > 99% accuracy, across all tasks, in both English and Hebrew.

In addition, at each time-point, the non-linguistic subtests of the Cognitive Linguistic Quick Test (CLQT; Helm-Estabrooks, 2001) were administered in order to assess whether non-linguistic cognitive skills remained stable across the study. For all cognitive skills assessed (attention, executive functions, visuospatial skills and clock drawing), scores were within-normal-limits at baseline and post-treatment relative to age. See Supplementary Table 1 for the specific CLQT scores.

Procedure: treatment

The participant received VNeST in Hebrew twice a week for 10 weeks, for about 1.5 hours per session. Overall, the participant received 30 hours of VNeST. Treatment was provided by two SLT students who were trained on the VNeST protocol and who each provided one

(1)

A *VERB* was presented to the participant, written in 1st person present tense, in Hebrew script, on an index card. The participant was asked to read the *VERB* aloud, then the card was removed, and the participant was asked to write down the *VERB*. If she struggled to write the *VERB* independently, the index card was returned, and the participant was asked to copy the *VERB*.

(2)

The participant was asked to produce four Subject-*VERB*-Object sentences. The participant was asked "Who *VERBs* what?" or "what might someone *VERB*?". If she was unable to produce agents and patients related to the *VERB* independently, she was offered a minimal cue (e.g., "Can you think of someone who *VERBs* something as part of their job?") or failing that, a maximal cue (a written choice of four options, one correct and three incorrect). As the participant produced each agent and patient, the SLT wrote down the responses on index cards. After producing each sentence verbally, the participant was asked to copy the whole sentence. After the four sentences were produced and written, the participant was asked to read them aloud.

(3)

The participant chose one sentence, answered WH-questions on that sentence (where, why, when), and then read aloud the expanded sentence.

(4)

The participant was asked to make plausibility judgements on twelve Subject-*VERB*-Object sentences read aloud by the SLT. Sentences that were not semantically feasible included those with an unrelated subject or object, or where the subject and object were reversed.

(5)

The participant was asked to recall the *VERB* both orally and orthographically.

(6)

The participant was asked to independently produce up to four Subject-*VERB*-Object sentences.

Figure 3. The six-stage Verb Network Strengthening Treatment protocol followed in this study, based closely on Edmonds (2014) protocol.

treatment session a week, in a quiet room at the participant's home or in the Hadassah Academic College SLT clinic. We note here that based on the current literature, both languages are expected to continue to decline as the PPA progresses, and therefore treating either language to slow the decline should be beneficial. These treatment effects could potentially also generalise to the untreated language. Although this was explained to the participant, she was only willing to work in Hebrew because her English was still functional, but her Hebrew was not. She rejected the option of a consecutive treatment block in English.

The VNeST protocol used for this study was based closely on Edmonds (2014) protocol, but with an added element of writing. Thus, for every verb trained (= 1 verb cycle), six stages were applied (see, Figure 3). Overall, 83 verb cycles were completed, taken from a pool of 20 verbs that were trained in a pseudo-random order. The 20 verbs were chosen specifically to contain no English-Hebrew cognates, and to share argument structure across the two languages, with half the trained verbs assessed in the sentence construction subtest of the REHAB and half not appearing in any subtest of the REHAB. See, Table 3

Table 3. List of verbs trained with VNeST during the treatment block.

Verbs that do not appear in the Revised English-Hebrew Aphasia Battery		Verbs that were assessed in the Revised English-Hebrew Aphasia Battery (Sentence Construction subtest)	
Hebrew	English translation	Hebrew	English translation
לאפות	To bake	לשקול	To weigh
להשאיל	To lend	לפתוח	To open
למצוא	To find	לבעוט	To kick
לעזוב	To leave	לתפוס	To catch
להיאבק	To fight	לשמור	To guard
לבחור	To examine	לעצור	To stop
לצפות	To watch	למזוג	To pour
לצעוק	To shout	לשתול	To plant
לחבק	To hug	לסחוב	To carry
לאבד	To lose	לשטוף	To wash

for the list of trained verbs. Treatment fidelity of our modified VNeST protocol was calculated to be > 95% based on 45% of treatment sessions that were observed live or via a video recording by the P.I.

Procedure: statistical analysis

We analysed whether learning occurred during treatment by charting retrieval abilities for every agent and patient produced within each sentence for every verb cycle throughout the treatment block. We then calculated correlations between treatment session and the average number of agents and patients retrieved per verb independently, after a minimal cue, or after a maximal cue during Stage 2 of the protocol (see, Figure 3). The maximum number of thematic roles per verb was eight – one agent and one patient per sentence across four sentences. Learning was accepted to have occurred during treatment if correlations were positive between treatment session and the average number of agents and patients retrieved independently per verb, and/or if correlations were negative between treatment session and the average number of agents and patients retrieved after a minimum or maximum cue.

We also analysed generalisation to other stimuli and other contexts in both languages using the REHAB and the WAB-R. Due to the degenerative nature of PPA, we looked at whether language skills were stable (or even improved) post-treatment compared to pre-treatment, or whether language skills continued to decline during the treatment period. In the REHAB, for subtests that allow individual stimuli to be compared across assessment time-points (e.g., object naming, action naming and non-word repetition) the McNemar test of Equal Change was calculated. No adjustment for multiple comparisons was necessary, because no McNemar calculation indicated significant change. For subtests that do not allow for individual stimuli to be compared across assessment time-points (due to more than one possible answer, e.g., sentence construction, WH-questions, oral narratives, written narratives), we calculated effect sizes using both Cohen's *d* and Non-Overlap of All Pairs (NAP), two measures that complement each other in that Cohen's *d* gives information about the size of change (Beeson & Robey, 2006) whereas NAP scores indicate how much overlap occurs between pre- and post-treatment data points (Conner et al., 2018; Parker & Vannest, 2009). When Cohen's *d* indicated a change of 1.2 or above

Table 4. Pre- and post-treatment scores for the REHAB and WAB-R in Hebrew (treated language) and English (untreated language).

		Hebrew			English		
		Pre	Post	Sig.	Pre	Post	Sig.
Object naming	Accuracy	60.0%*	57.78%	McN. = 0.11	97.78%	95.56%	McN. = 1
Action naming	Accuracy	26.67%*	22.22%	McN. = 0.5	82.22%	86.67%	McN. = 0.14
Sentence construction	Rel. SVO production accuracy	50.0%	53.70%	ES(*d*) = 0.58 NAP = .33	90.74%	94.44%	ES(*d*) = 0.67 NAP = .22
Answering WH-questions	Full and appropriate answer	87.50%	100%	ES(*d*) = 1.15 NAP = .56	100%	100%	n/a
Oral narratives	Rel. SVO utterances out of total utterances (=%CUs)	**52.0%**	**38.26%**	**ES(*d*) = −1.90 NAP = −.78**	**89.68%**	**79.31%**	**ES(*d*) = −1.66 NAP = −1.0**
	Type/token ratios – noun	**0.84**	**0.70**	**ES(*d*) = −2.06 NAP = −.78**	**0.80**	**0.75**	**ES(*d*) = −1.62 NAP = −.78**
	Type/token ratios – verb	0.70	0.76	ES(*d*) = 0.64 NAP = .56	0.74	0.71	ES(*d*) = −0.61 NAP = −.33
Written narratives	Rel. SVO sentences out of total sentences (=%CSs)	61.90%	65.0%	ES(*d*) = 0.18 NAP = 0	95.65%	82.35%	ES(*d*) = −1.15 NAP = −0.11
	Type/token ratios – noun	0.71	0.77	ES(*d*) = 0.52 NAP = .33	0.90	0.90	ES(*d*) = −0.13 NAP = .11
	Type/token ratios – verb	0.73	0.75	ES(*d*) = 0.50 NAP = .33	0.81	0.72	ES(*d*) = −0.89 NAP = −.44
Non-word repetition	Accuracy	56.67%	63.33%	McN. = 0.33	70.0%	66.67%	McN. = 0.08
WAB-R	AQ (/100)	60.2	53.5	n/a	91.4	86.8	n/a
	Spontaneous speech (/20)	13	12	n/a	19	19	n/a
	Auditory verbal (/10)	9	8.45	n/a	9.8	8.7	n/a
	Repetition (/10)	3.4	2.6	n/a	7.6	6.7	n/a
	Naming and word finding (/10)	4.7	3.7	n/a	9.3	9	n/a

Note. Pre = pre-treatment; post = post-treatment; sig. = significance; CUs = Complete Utterances; CSs = Complete Sentences; Rel. = relevant; REHAB = Revised English-Hebrew Aphasia Battery; WAB-R = Western Aphasia Battery Revised; McN. =McNemar test of equal change; ES (*d*) = Effect size as measured by Cohen's *d*; NAP = Effect size as measured by Non-overlap of All Pairs. Bold font indicates observed decline.

*Significant difference between baseline object and action naming in Hebrew; t = 2.13, p = .004

ǂSignificant difference between baseline object and action naming in English; t = 2.13, p = .048

(Edmonds, 2014), together with a medium or high NAP score (.32–1.0), change was assumed to have occurred – positive scores indicated improvement, and negative scores indicated decline.

For the WAB-R, we did not have multiple baselines, so we looked at (1) a change of more than 5-points, which may indicate functional change (Katz & Wertz, 1997), and (2) patterns of decline and/or stability relative to the participant's decline in the five years leading up to the study.

Results

Learning during treatment was observed: as sessions progressed, the average independent retrieval of thematic roles per verb increased significantly ($r(20) = .552$, $p = .011$), whereas the average retrieval after a minimal cue decreased significantly ($r(20) = .566$, $p = .009$). Retrieval of a thematic role after a maximal cue occurred just once, during the first session, and remained at zero for the remainder of the sessions. We thus accepted that treatment effects, as measured by within- and cross-language generalisation, could be attributed to VNeST.

Within-language generalisation measures of the treated language indicated that the participant's WAB-R AQ decreased in Hebrew, from 60.2 (pre-treatment) to 53.5 (post-treatment), a decrease of more than 5-points that could indicate a functional decline (Katz & Wertz, 1997). This decline was observed mainly in the subtests of spontaneous speech, repetition (of words and sentences), and naming and word-finding (see, Figures 1 and 2, from July to October 2019). However, notwithstanding this continued observed decline in the WAB-R, no significant decline was observed in the REHAB for lexical production in word or sentence level tasks, or for written narratives. Conversely, for oral narratives, decline was observed for the percentage of Complete Utterances (relevant SVO sentences) and noun diversity (as measured by the noun type-token ratio), while verb diversity (as measured by the verb type-token ratio) remained stable. We also note stable non-word repetition skills in Hebrew (i.e., non-words based on Hebrew phonology). See, Table 4 for pre- and post-treatment Hebrew language measures.

Cross-language measures of the untreated language indicated that the participant's WAB-R AQ decreased in English, from 91.4 (pre-treatment) to 86.8 (post-treatment). This decrease of less than 5-points potentially indicates no functional decline (Katz & Wertz, 1997), but with the absence of multiple baselines we were unable to conclude whether decline was significant or not, particularly since the decrease was close to 5-points. Interestingly, this continued trend of language decline based on the WAB-R was observed most saliently in the subtests of repetition (words and sentences), and auditory-verbal comprehension, but not for spontaneous speech or naming and word-finding (see, Figures 1 and 2, from July to October 2019). Similar to Hebrew skills, in the REHAB in English no significant decline was observed for lexical production in word or sentence level tasks, or for written narratives. Furthermore, similar to Hebrew skills, for oral narratives in English a decline was observed for the percentage of Complete Sentences (relevant SVO sentences) and noun diversity, while verb diversity remained stable. Here too, we note stable non-word repetition skills in English (i.e., non-words based on English phonology). See, Table 4 for pre- and post-treatment English language measures.

Discussion

In our study, we examined the lexical retrieval skills of an English-Hebrew bilingual with lvPPA post-treatment relative to pre-treatment. Our data supported the possibility that VNeST may have interrupted the decline of lexical retrieval impairment of single-words and sentences in both the treated and untreated languages during the study, relative to a continued general decline in language skills, although the results were not conclusive. Our findings also partially paralleled those of Meyer et al. (2015) in their pioneering treatment study of a bilingual person with lvPPA, even though treatment protocol, timeline, and assessment measures were different.

We first asked whether providing VNeST in the post-morbidly more-impaired language of a bilingual individual with lvPPA would interrupt the deterioration of lexical retrieval in that language. We found that lexical retrieval in single-word and sentence-level tasks, as well as in written narratives, did not decline when tested post-treatment with the REHAB compared to pre-treatment (Berthier & Pulvermüller, 2011; Fox & Stryker, 2017; Meyer et al., 2020). Thus, our results provide evidence for the efficacy of the treatment (Edmonds et al., 2009) and support our hypothesis that lexical retrieval would remain stable in the treated language, which we attribute to a strengthened semantic network.

Conversely, in the Hebrew WAB-R, we found that lexical retrieval skills appeared to continue to decline in the naming and word-finding subtest post-treatment compared to pre-treatment. However, a closer look at this subtest showed that for object naming, accuracy was actually higher in Hebrew post-treatment compared to pre-treatment but pre-treatment most of her errors were correct responses in the non-target language (immediately, without attempting to name in Hebrew), whereas post-treatment she named most of the objects correctly but after a phonemic cue (in Hebrew). Thus, her subtest score was greatly reduced post-treatment due to cueing. For sentence completion, she improved and for responsive speech she declined but crucially her errors in both tasks were all in Hebrew (semantic paraphasias) post-treatment, whereas pre-treatment her errors were all correct responses but in the non-target language. This pattern of errors supports our hypothesis that a strengthened semantic network results in better lexico-semantic connections and thus better access to Hebrew, especially since phonological cueing should have reduced the phonological load and aided retrieval in Hebrew. Alternatively, the results could indicate that interference from her less-impaired English was controlled better due to the Hebrew treatment. However due to her 90% daily use of English outside of the Hebrew treatment block, this explanation seems less likely because she was not using Hebrew primarily during the 10 weeks of treatment generally, and thus was not consistently practicing controlling interference from English. A further possibility is that the post-treatment increase in attempting to name in Hebrew indicates improved confidence after the Hebrew treatment block.

We further note that the differential results observed in the REHAB and the WAB-R for naming could potentially be due to different tasks (picture-naming and object-naming respectively), and/or different psycholinguistic properties of the words, such as frequency, word length etc., which were controlled for within groups and across languages in the REHAB but not between the WAB-R and the REHAB. However, we have no indication that the participant found naming objects (in the WAB-R) harder than naming pictures (in the REHAB), or that the words in the REHAB were intrinsically easier than those in the WAB-R from a psycholinguistic perspective, based on the participant's high and comparable scores in both noun-naming subtests in English. We accept, however, that we did not have multiple baselines for the WAB-R, and so the continued trend of decline observed may not have been interpreted as change based on effect sizes such as Cohen's *d* or NAP, as we were able to calculate for the REHAB. These factors warrant further investigation, together with the use of error analyses to support quantitative measures.

One hypothesis that might explain the lack of decline observed in the REHAB is that due to the relatively short time between pre-treatment and post-treatment assessment, no further decline occurred due to PPA. However, our data do not support this hypothesis for the following reasons. First, we found that Hebrew lexical retrieval skills declined in oral narratives in the REHAB over the 10-week treatment block. Second, we found a continued decline in the Hebrew WAB-R post-treatment relative to the five years before our study, as observed in the AQ as well as in each subtest (although minimally in auditory-verbal comprehension). While we have explained possible reasons for the apparent decline in naming and word-finding above, abilities continued to decline in Hebrew for repetition and for word-finding in oral narratives. These tasks were not specifically targeted during treatment, and often decline in lvPPA (Gorno-Tempini et al., 2011; Mesulam, 2007), notably in those with temporoparietal hypoperfusion, as in our participant (Krishnan et al., 2016), due to the strong reliance on cognitive processes that are impaired, such as auditory-verbal working memory, reduced mental flexibility, and fluency (e.g., Krishnan et al., 2016; Neerincx, 2003; Shadden et al., 1991).

Even though our participant's non-linguistic cognitive skills were within normal limits, it is plausible that these same cognitive skills, when engaged in a language task, would be saliently reduced. This may explain why the repetition task and lexical retrieval during oral narratives continued to decline. Furthermore, the stable lexical retrieval skills observed for written narratives support this hypothesis, because written narratives rely less on cognitive processes such as auditory-verbal working memory, there is more time to respond, and self-monitoring occurs during and after writing. However, the stable written narrative skills observed may also be related to the different language pathways involved in orthographical output vs. phonological output.

Indeed, Meyer et al. (2015) suggested that the results from their study, which included generalisation to untrained lexical stimuli in the treated language (L2) at the single-word level after orthographic treatment but no generalisation after phonological treatment, indicated that the semantic network was more involved in orthographic treatment than in phonological treatment. However, no explanation as to why the authors thought there was a differential contribution of semantics to the different treatment types was offered. Furthermore, they provided both treatments simultaneously (albeit with different stimuli). Future research is needed to analyse the effects of different types of treatment on different language tasks, to better understand the contribution of cognitive impairments resulting from lvPPA and their engagement during language tasks.

Our second research question referred to whether any treatment effects generalised to the untreated language. We found some indication that generalisation may have occurred because we observed similar effects in English as in Hebrew for the subtests of the REHAB, with no significant decline for lexical retrieval skills in single-word naming, sentence-level tasks, and written narratives. Thus, our results are partially aligned with our hypothesis in that we expected generalisation of treatment effects to the untreated language due to shared semantic knowledge and overlapping neural substrates subserving that knowledge (Paradis, 1993).

We acknowledge that the lack of decline in English for lexical retrieval skills in single-word naming, sentence-level tasks, and written narratives may have been due to other factors, such as the short time-frame of the study, during which decline in the less-impaired language may not have occurred (potentially more likely than in Hebrew, due to the slower decline generally in English). Likewise, during the study, our participant reported producing English 90% of the day, compared to 10% in Hebrew. Thus, her English production skills may have remained stable due to daily use rather than specific treatment effects. However, due to the oral narrative data in English that significantly declined post-treatment relative to pre-treatment, similar to Hebrew, we cautiously interpret our data as showing possible generalisation effects from treatment.

Further support comes from the WAB-R in English which continued to decline post-treatment relative to the previous 5-years (where decline was observed almost exclusively in the repetition subtest). While the decline was smaller than in Hebrew and potentially not significant, notably, repetition continued to decline, as well as auditory-verbal comprehension. While decline in repetition is expected in lvPPA, due to damage to the phonological network resulting in reduced auditory-verbal working memory skills (e.g., Krishnan et al., 2016), auditory-verbal comprehension is relatively spared in the early stages of the disease (Gorno-Tempini et al., 2011; Mesulam, 2007). However, as the disease progresses, decline in sentence comprehension may become more salient due to deficits in auditory-verbal working memory. Indeed, our participant only showed decline for complex sentence comprehension in the WAB-R, but not for comprehension of words or simple sentences. It may be that this decline in English occurred as the L1 deteriorated and closed the gap with the L2, a pattern of decline in bilingual people with PPA that has been observed previously (Costa et al., 2019; Malcolm et al., 2019) but which perhaps was not observed for the spontaneous speech or naming and word-finding subtests due to treatment effects. While VNeST includes an element of auditory comprehension, it remains at the simple-sentence level; future research into different types of treatment and their effects on more specific comprehension and production measures should help to understand the observed pattern of decline.

Our results are similar to those of Meyer et al. (2015), who also found some indication of cross-language generalisation with orthographic treatment effects generalising to the untreated language more saliently than phonological treatment. As mentioned previously, the researchers suggested that this indicated a more strongly engaged semantic network during orthographic treatment than during phonological treatment (Meyer et al., 2015). Our data support this hypothesis, because we potentially also found generalisation to the untreated language after a semantic-based treatment (e.g., Kroll & Tokowicz, 2005; Meyer et al., 2015), which we attribute to strengthening shared semantic knowledge across languages (Paradis, 1993).

Limitations

To our knowledge, this is only the second study on treatment of lvPPA in a bilingual person, and the results encouragingly support the previous study (Meyer et al., 2015) despite the methodological differences. However, there are several limitations to this study that must be considered when interpreting the results and planning future research. First, the participant received 10 weeks of treatment but following the post-treatment assessments did not agree to continue to receive more treatment or be assessed after a period of no treatment (to assess maintenance and/or further decline). This resulted in a shorter study than originally anticipated, and thus observing decline over time was challenging because there may not have been decline in certain language skills over just 10 weeks – PPA does not necessarily decline in a linear manner over time (Gorno-Tempini et al., 2011). However, by comparing different language skills in different contexts, we found support that the observed stable lexical retrieval skills were potentially due to treatment effects, relative to other skills and/or contexts where language abilities continued to decline.

A second limitation can be found in our analyses of the different language tasks. We were able to conduct statistical analyses on the data from the REHAB, due to multiple baselines that we collected in each language without excessive repetitiveness because of the split battery (Borodkin et al., 2020). Nevertheless, due to long and somewhat fatiguing testing sessions we only collected data from the WAB-R once (in each language) at each testing time-point, and thus were unable to conduct statistical analyses on that data. Furthermore, the WAB-R data recorded in the participant's medical file was collected at only three time-points in English and twice in Hebrew prior to this study (during a period of 5-years). We were therefore unable to conduct statistical analyses on this sparse data and accept that when we discuss a trend of decline in subtests of the WAB-R, there may be no significant difference pre- to post-treatment compared to the previous 5-years.

A third limitation is that we did not compare different types of treatment (e.g., semantic-focused treatment compared to phonological-focused treatment), rather, we investigated the effect of one treatment – VNeST – on lexical retrieval skills. We expected deficits in the functioning of the phonological network, or access to it, due to the lvPPA diagnosis, but it was unclear whether treatment effects (potentially due to a strengthened semantic network) boosted lexical retrieval generally, or specifically as a response to a weakened semantic network due to atrophy spreading along the temporal pole (Gorno-Tempini et al., 2011; Mesulam, 2007). Future research should focus on different treatment types and their effect on specific semantic and phonological language measures, relative to different subtypes of PPA and their underlying pathology.

Conclusion

In summary, our case study on a bilingual participant with lvPPA offers important yet inconclusive information on treatment for lvPPA in multilingual people. Our results indicate that VNeST may be an effective prophylactic treatment for lexical retrieval skills in both treated and untreated languages in this population. More studies are necessary to identify which treatments are most effective, for which types of PPA, in which languages (the treated language/s or also the untreated language/s), and over what time frame.

Continued research into treatment of PPA in multilingual people will provide crucial information regarding effective treatment moving forward in this field.

Acknowledgments

We thank the participant in the study for her time and commitment to this research. We also thank our three research assistants, Anna Wolfbein, Gal Lavi and Ori Menachem who, together with the third author (Y.N.) collected and transcribed the data and/or provided treatment to the participant. Furthermore, we are grateful to the anonymous reviewers who provided helpful and important feedback on earlier drafts of this paper.

Disclosure statement

No potential conflict of interest was reported by the author(s).

Funding

The author(s) reported there is no funding associated with the work featured in this article.

References

Bastiaanse, R., Edwards, S., & Rispens, J. (2002). *Verb and Sentence test (VAST)*. Suffolk: Thames Valley Test Company.

Beales, A., Cartwright, J., Whitworth, A., & Panegyres, P. K., et al. (2016). Exploring generalisation processes following lexical retrieval intervention in primary progressive aphasia. *International Journal of Speech-Language Pathology*, *18*(3), 299–314. https://doi.org/10.3109/17549507.2016.1151936

Beeson, P. M., King, R. M., Bonakdarpour, B., Henry, M. L., Cho, H., & Rapcsak, S. Z., et al. (2011). Positive effects of language treatment for the logopenic variant of primary progressive aphasia. *Journal of Molecular Neuroscience*, *45*(3), 724–736. https://doi.org/10.1007/s12031-011-9579-2

Beeson, P. M., & Robey, R. R. (2006). Evaluating single-subject treatment research: lessons learned from the aphasia literature. *Neuropsychology Review*, *16*(4), 161–169. https://doi.org/10.1007/s11065-006-9013-7

Berthier, M. L., & Pulvermüller, F. (2011). Neuroscience insights improve neurorehabilitation of poststroke aphasia. *Nature Reviews. Neurology*, *7*(2), 86–97. https://doi.org/10.1038/nrneurol.2010.201

Borodkin, K., Goral, M., & Kempler, D., et al. (2020). Measuring performance stability in persons with aphasia: identical versus comparable testing forms. *Aphasiology*, *34*(3), 376–390. https://doi.org/10.1080/02687038.2019.1670331

Cadório, I., Lousada, M., Martins, P., & Figueiredo, D., et al. (2017). Generalization and maintenance of treatment gains in primary progressive aphasia (PPA): A systematic review. *International Journal of Language & Communication Disorders*, *52*(5), 543–560. https://doi.org/10.1111/1460-6984.12310

Conner, P. S., Goral, M., Anema, I., Borodkin, K., Haendler, Y., Knoph, M., Mustelier, C., Paluska, E., Melnikova, Y., & Moeyaert, M., et al. (2018). The role of language proficiency and linguistic distance in cross-linguistic treatment effects in aphasia. *Clinical Linguistics & Phonetics*, *32*(8), 739–757. https://doi.org/10.1080/02699206.2018.1435723

Costa, A. S., Jokel, R., Villarejo, A., Llamas-Velasco, S., Domoto-Reilley, K., Wojtala, J., Reetz, K., & Machado, Á., et al. (2019). Bilingualism in primary progressive aphasia: A retrospective study on clinical and language characteristics. *Alzheimer Disease and Associated Disorders*, *33*(1), 47–53. https://doi.org/10.1097/WAD.0000000000000288

Druks, J., & Weekes, B. S. (2013). Parallel deterioration to language processing in a bilingual speaker. *Cognitive Neuropsychology*, *30*(7–8), 578–596. https://doi.org/10.1080/02643294.2014.882814
Edmonds, L. A., & Babb, M. (2011). Effect of verb network strengthening treatment in moderate-to-severe aphasia. *American Journal of Speech-Language Pathology*, *20*(2), 131. https://doi.org/10.1044/1058-0360(2011/10-0036)
Edmonds, L. A., Nadeau, S. E., & Kiran, S., et al. (2009). Effect of verb network strengthening treatment (VNeST) on lexical retrieval of content words in sentences in persons with aphasia. *Aphasiology*, *23*(3), 402–424. https://doi.org/10.1080/02687030802291339
Edmonds, L. A. (2014). Tutorial for verb network strengthening treatment (VNeST): detailed description of the treatment protocol with corresponding theoretical rationale. *Perspectives on Neurophysiology and Neurogenic Speech and Language Disorders*, *24*(3), 78. https://doi.org/10.1044/nnsld24.3.78
Edmonds, L. A. (2016). A review of verb network strengthening treatment: theory, methods, results, and clinical implications. *Topics in Language Disorders*, *36*(2), 123–135. https://doi.org/10.1097/TLD.0000000000000088
Fabbro, F. (1999). Aphasia in multilinguals. In F. Fabbro (Ed.), *Concise encyclopedia of language pathology* (pp. 335–341). Pergamon Press.
Filley, C. M., Ramsberger, G., Menn, L., Wu, J., Reid, B. Y., & Reid, A. L., et al. (2006). Primary progressive aphasia in a bilingual woman. *Neurocase*, *12*(5), 296–299. https://doi.org/10.1080/13554790601126047
Fox, K., & Stryker, M. (2017). Integrating hebbian and homeostatic plasticity: introduction. *Philosophical Transactions of the Royal Society B: Biological Sciences*, *372*(1715), 20160413. https://doi.org/10.1098/rstb.2016.0413
García-Pentón, L., Pérez Fernández, A., Iturria-Medina, Y., Gillon-Dowens, M., & Carreiras, M., et al. (2014). Anatomical connectivity changes in the bilingual brain. *NeuroImage*, *84*, 495–504. https://doi.org/10.1016/j.neuroimage.2013.08.064
Gil, M., & Edelstein, T. (2001). *Hebrew version of the PALPA: Psycholinguistic assessment of language processing*. Raanana, Israel: Lowenstein Rehabilitation Center.
Gollan, T. H., Weissberger, G. H., Runnqvist, E., Montoya, R. I., & Cera, C. M. (2012). Self-ratings of spoken language dominance: A Multilingual Naming Test (MINT) and preliminary norms for young and aging Spanish–English bilinguals. *Bilingualism: Language and Cognition*, *15*(03), 594–615. https://doi.org/10.1017/S1366728911000332
Goral, M., & Lerman, A. (2020). Variables and mechanisms affecting response to language treatment in multilingual people with aphasia. *Behavioral Sciences*, *10*(9), 144. https://doi.org/10.3390/bs10090144
Gorno-Tempini, M. L., Brambati, S. M., Ginex, V., Ogar, J., Dronkers, N. F., Marcone, A., & Miller, B. L., et al. (2008). The logopenic/phonological variant of primary progressive aphasia. *Neurology*, *71*(16), 1227–1234. https://doi.org/10.1212/01.wnl.0000320506.79811.da
Gorno-Tempini, M. L., Hillis, A. E., Weintraub, S., Kertesz, A., Mendez, M., Cappa, S. F., Ogar, J. M., Rohrer, J. D., Black, S., Boeve, B. F., Manes, F., Dronkers, N. F., Vandenberghe, R., Rascovsky, K., Patterson, K., Miller, B. L., Knopman, D. S., Hodges, J. R., Mesulam, M. M., & Grossman, M., et al. (2011). Classification of primary progressive aphasia and its variants. *Neurology*, *76*(11), 1006–1014. https://doi.org/10.1212/WNL.0b013e31821103e6
Halsband, U. (2006). Bilingual and multilingual language processing. *Journal of Physiology-Paris*, *99*(4–6), 355–369. https://doi.org/10.1016/j.jphysparis.2006.03.016
Helm-Estabrooks, N. (2001). *Cognitive Linguistic Quick Test*. The Psychological Corporation.
Henry, M. L., Hubbard, H. I., Grasso, S. M., Dial, H. R., Beeson, P. M., Miller, B. L., & Gorno-Tempini, M. L., et al. (2019). Treatment for word retrieval in semantic and logopenic variants of primary progressive aphasia: immediate and long-term outcomes. *Journal of Speech, Language, and Hearing Research*, *62*(8), 2723–2749. https://doi.org/10.1044/2018_JSLHR-L-18-0144
Henry, M. L., Rising, K., DeMarco, A. T., Miller, B. L., Gorno-Tempini, M. L., & Beeson, P. M., et al. (2013). Examining the value of lexical retrieval treatment in primary progressive aphasia: two positive cases. *Brain and Language*, *127*(2), 145–156. https://doi.org/10.1016/j.bandl.2013.05.018

Katz, R. C., & Wertz, R. T. (1997). The efficacy of computer provided reading treatment for chronic aphasic adults. *Journal of Speech, Language, and Hearing Research*, *40*(3), 493–507. https://doi.org/10.1044/jslhr.4003.493

Kertesz, A. (2006). *Western Aphasia Battery—Revised*. Pro-Ed.

Kiran, S., & Bassetto, G. (2008). Evaluating the effectiveness of semantic-based treatment for naming deficits in aphasia: what works? *Seminars in speech and language*, *29*, 071–082. Thieme Medical Publishers. https://doi.org/10.1055/s-2008-1061626

Kiran, S., Sandberg, C., Gray, T., Ascenso, E., & Kester, E., et al. (2013). Rehabilitation in bilingual aphasia: evidence for within- and between-language generalization. *American Journal of Speech-Language Pathology*, *22*(2), S298. https://doi.org/10.1044/1058-0360(2013/12-0085

Krishnan, K., Machulda, M. M., Whitwell, J. L., Butts, A. M., Duffy, J. R., Strand, E. A., Senjem, M. L., Spychalla, A. J., Jack, C. R., Lowe, V. J., & Josephs, K. A., et al. (2016). Varying degrees of temporoparietal hypometabolism on FDG-PET reveal amyloid-positive logopenic primary progressive aphasia is not a homogeneous clinical entity. *Journal of Alzheimer's Disease*, *55*(3), 1019–1029. https://doi.org/10.3233/JAD-160614

Kroll, J. F., & Tokowicz, N. (2005). Models of bilingual representation and processing: Looking back and to the future. In J. F. Kroll & A. M. B. de Groot (Eds.), *Handbook of bilingualism: Psycholinguistic approaches* (pp. 531–554). Oxford University Press.

Kroll, J. F., Van Hell, J. G., Tokowicz, N., & Green, D. W., et al. (2010). The revised hierarchical model: A critical review and assessment. *Bilingualism: Language and Cognition*, *13*(3), 373–381. https://doi.org/10.1017/S136672891000009X

Laine, M., & Martin, N. (2006). *Anomia: Clinical and theoretical aspects*. Psychology Press.

Larner, A. J. (2012). Progressive non-fluent aphasia in a bilingual subject: relative preservation of "Mother Tongue". *Journal of Neuropsychiatry and Clinical Neuroscience*, *24*(1), E9–E10. https://doi.org/10.1176/appi.neuropsych.11010019

Lerman, A., Edmonds, L. A., & Goral, M., et al. (2018). Cross-language generalisation in bilingual aphasia: what are we missing when we do not analyse discourse? *Aphasiology*, *33*(9), 1154–1162. https://doi.org/10.1080/02687038.2018.1538493

Lerman, A., Goral, M., Edmonds, L. A., & Obler, L. K., et al. (in press). Strengthening the semantic verb network in multilingual people with aphasia: Within- and cross-language treatment effects. *Bilingualism: Language and Cognition*. https://doi.org/ 10.1017/S1366728921001036 .

Lerman, A., & Goral, M. (unpublished). *Revised English-Hebrew Aphasia Battery*.

Liu, Y. C., Yip, P. K., Fan, Y. M., & Meguro, K., et al. (2012). A potential protective effect in multilingual patients with semantic dementia: Two case reports of patients speaking Taiwanese and Japanese. *Acta Neurol Taiwan*, *21*(1), 25–30.

Machado, A., Rodrigues, M., Simoes, S., & Santana, I., et al. (2010). The Portuguese who could no longer speak french: primary progressive aphasia in a bilingual man. *Journal of Neuropsychiatry and Clinical Neuroscience*, *22*(1), E31–E32. https://doi.org/10.1176/jnp.2010.22.1.123.e31

Malcolm, T. R., Lerman, A., Korytkowska, M., Vonk, J. M. J., & Obler, L. K., et al. (2019). Primary progressive aphasia in bilinguals and multilinguals. In J. Schweiter (Ed.), *The handbook of the neuroscience of multilingualism* (pp. 572–591). Wiley.

Marian, V., Spivey, M., & Hirsch, J., et al. (2003). Shared and separate systems in bilingual language processing: converging evidence from eyetracking and brain imaging. *Brain and Language*, *86*(1), 70–82. https://doi.org/10.1016/S0093-934X(02)00535-7

Mesulam, M. M., Rogalski, E. J., Wieneke, C., Hurley, R. S., Geula, C., Bigio, E. H., Thompson, C. K., & Weintraub, S., et al. (2014). Primary progressive aphasia and the evolving neurology of the language network. *Nature Reviews. Neurology*, *10*(10), 554–569. https://doi.org/10.1038/nrneurol.2014.159

Mesulam, M. M. (2001). Primary progressive aphasia. *Annals of Neurology*, *49*(4), 425–432. https://doi.org/10.1002/ana.91

Mesulam, M. M. (2007). Primary progressive aphasia: A 25-year retrospective. *Alzheimer Disease and Associated Disorders*, *21*(4), S8–S11. https://doi.org/10.1097/WAD.0b013e31815bf7e1

Meyer, A. M., Getz, H. R., Brennan, D. M., Hu, T. M., & Friedman, R. B., et al. (2016). Telerehabilitation of anomia in primary progressive aphasia. *Aphasiology*, *30*(4), 483–507. https://doi.org/10.1080/02687038.2015.1081142

Meyer, A. M., Snider, S. F., Eckmann, C. B., & Friedman, R. B., et al. (2015). Prophylactic treatments for anomia in the logopenic variant of primary progressive aphasia: cross-language transfer. *Aphasiology*, *29*(9), 1062–1081. https://doi.org/10.1080/02687038.2015.1028327

Meyer, A. M., Snider, S. F., McGowan, S. A., Tippett, D. C., Hillis, A. E., & Friedman, R. B., et al. (2020). Grammatical ability predicts relative action naming impairment in primary progressive aphasia. *Aphasiology*, *34*(6), 664–674. https://doi.org/10.1080/02687038.2020.1734527

Meyer, A. M., Tippett, D. C., & Friedman, R. B., et al. (2018). Prophylaxis and remediation of anomia in the semantic and logopenic variants of primary progressive aphasia. *Neuropsychological Rehabilitation*, *28*(3), 352–368. https://doi.org/10.1080/09602011.2016.1148619

Nadeau, S. E. (2019). Bilingual aphasia: explanations in population encoding. *Journal of Neurolinguistics*, *49*, 117–143. https://doi.org/10.1016/j.jneuroling.2018.10.002

Neerincx, M. A. (2003). Cognitive task load analysis: Allocating tasks and designing support. In E. Hollnagel (Ed.), *Handbook of cognitive task design* (pp. 283–305). Lawrence Erlbaum.

Newhart, M., Davis, C., Kannan, V., Heidler-Gary, J., Cloutman, L., & Hillis, A. E., et al. (2009). Therapy for naming deficits in two variants of primary progressive aphasia. *Aphasiology*, *23*(7–8), 823–834. https://doi.org/10.1080/02687030802661762

Nickels, L. (2002). Therapy for naming disorders: revisiting, revising, and reviewing. *Aphasiology*, *16* (10–11), 935–979. https://doi.org/10.1080/02687030244000563

Paradis, M. (2011). Principles underlying the Bilingual Aphasia Test (BAT) and its uses. *Clinical Linguistics & Phonetics*, *25*(6–7), 427–443. https://doi.org/10.3109/02699206.2011.560326

Paradis, M. (1993). Bilingual aphasia rehabilitation. In M. Paradis (Ed.), *Foundations of aphasia rehabilitation* (pp. 413–419). Pergamon Press.

Parker, R. I., & Vannest, K. (2009). An improved effect size for single-case research: nonoverlap of all pairs. *Behavior Therapy*, *40*(4), 357–367. https://doi.org/10.1016/j.beth.2008.10.006

Rapp, B., & Glucroft, B. (2009). The benefits and protective effects of behavioural treatment for dysgraphia in a case of primary progressive aphasia. *Aphasiology*, *23*(2), 236–265. https://doi.org/10.1080/02687030801943054

Raymer, A. M., & Ellsworth, T. A. (2002). Response to contrasting verb retrieval treatments: A case study. *Aphasiology*, *16*(10–11), 1031–1045. https://doi.org/10.1080/026870401430000609

Shadden, B. B., Burnette, R. B., Eikenberry, B. R., & DiBrezzo, R., et al. (1991). All discourse tasks are not created equal. *Clinical Aphasiology*, *20*, 327–342.

Soroker, N. (1997). *Hebrew Western Aphasia Battery*. Loewenstein Hospital Rehabilitation Center.

Tippett, D. C., Hillis, A. E., & Tsapkini, K., et al. (2015). Treatment of primary progressive aphasia. *Current Treatment Options in Neurology*, *17*(8), 1–11. https://doi.org/10.1007/s11940-015-0362-5

Van de Putte, E., De Baene, W., Price, C. J., & Duyck, W., et al. (2018). Neural overlap of L1 and L2 semantic representations across visual and auditory modalities: A decoding approach. *Neuropsychologia*, *113*, 68–77. https://doi.org/10.1016/j.neuropsychologia.2018.03.037

Van Hell, J. G., & De Groot, A. M. B. (1998). Conceptual representation in bilingual memory: effects of concreteness and cognate status in word association. *Bilingualism: Language and Cognition*, *1*(3), 193–211. https://doi.org/10.1017/S1366728998000352

Videsott, G., Herrnberger, B., Hoenig, K., Schilly, E., Grothe, J., Wiater, W., Spitzer, M., & Kiefer, M., et al. (2010). Speaking in multiple languages: neural correlates of language proficiency in multilingual word production. *Brain and Language*, *113*(3), 103–112. https://doi.org/10.1016/j.bandl.2010.01.006

Wambaugh, J. L., Doyle, P. J., Martinez, A. L., & Kalinyak-Fliszar, M., et al. (2002). Effects of two lexical retrieval cueing treatments on action naming in aphasia. *Journal of Rehabilitation Research and Development*, *39*(4), 4455–4466.

Westbury, C., & Bub, D. (1997). Primary progressive aphasia: A review of 112 cases. *Brain and Language*, *60*(3), 381–406. https://doi.org/10.1006/brln.1997.1840

Win, K. T., Pluta, J., Yushkevich, P., Irwin, D. J., McMillan, C. T., Rascovsky, K., & Grossman, M., et al. (2017). Neural correlates of verbal episodic memory and lexical retrieval in logopenic variant primary progressive aphasia. *Frontiers in Neuroscience*, *11*, 330. https://doi.org/10.3389/fnins.2017.00330

Wong, B., Yin, B., & O'Brien, B., et al. (2016). Neurolinguistics: structure, function, and connectivity in the bilingual brain. *BioMed Research International*, *2016*, 1–22. https://doi.org/10.1155/2016/7069274

Yang, Y., Wang, J., Bailer, C., Cherkassky, V., & Just, M. A., et al. (2017). Commonalities and differences in the neural representations of english, Portuguese, and mandarin sentences: when knowledge of the brain-language mappings for two languages is better than one. *Brain and Language*, *175*, 77–85. https://doi.org/10.1016/j.bandl.2017.09.007