School of Occupational Therapy, Social Work and Speech Pathology

Making the Right Connections: Assessment and Treatment of Lexical Retrieval Difficulties in People with Primary Progressive Aphasia and Alzheimer's Disease

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This thesis is presented for the Degree of
Doctor of Philosophy
of
Curtin University

July, 2020

Declaration

To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgment has been made. This thesis contains no material which has been accepted for the award of any other degree or diploma in any university.

The research presented and reported in this thesis was conducted in accordance with the National Health and Medical Research Council National Statement on Ethical Conduct in Human Research (2007) – updated March 2014. The proposed research study received human research ethics approval from the Curtin University Human Research Ethics Committee (EC00262), Approval Number #HR218/2015.

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Abstract

Lexical retrieval difficulties are a common symptom of language impairment in neurodegenerative conditions and are particularly prevalent in conditions such as primary progressive aphasia and Alzheimer's disease. These difficulties can have an overwhelming impact on communication and contribute to quality of social interactions and connectedness, ultimately impacting the quality of life of people with dementia. To date, speech-language pathology interventions for primary progressive aphasia and Alzheimer's disease have largely focused on remediation of lexical retrieval with positive treatment effects increasingly reported for treated words. Despite these promising treatment effects, intervention ingredients and outcome measures for assessment and optimisation of generalisation, respectively, are yet to be understood. Moreover, evaluation of such interventions from the perspectives of people with dementia and their family members is yet to be examined, a current barrier to future therapeutic endeavours.

This program of research aimed to provide insights into diagnosis, assessment, and intervention by seeking to deepen understanding of lexical retrieval impairment in people with primary progressive aphasia and Alzheimer's disease through five lines of enquiry that have culminated in five studies (four have been published and one submitted and currently under review). Each of these five strands, organised within three phases of research, build on each other to explore how intervention may be maximally effective and facilitate optimum generalisation of lexical gains in progressive language disorders. Following a purposeful review of the literature that aimed to examine the mechanisms of change underlying lexical retrieval intervention, a greater understanding emerged of how interventions had been designed and their respective outcomes, and areas of need identified that informed the following investigations. Through the subsequent detailed profiling of cognitive and language behaviours in people with different dementia syndromes, supported by neuroimaging data, and the design, implementation, and evaluation of a novel intervention protocol, this thesis has sought to both inform understanding of the nature of the impairments and advance clinical practice with these populations.

A mixed methods research design incorporating quantitative and qualitative data (diagnostic group and individual) were utilised in this research program. Twelve participants (four semantic variant primary progressive aphasia, four logopenic

variant primary progressive aphasia, and four Alzheimer's disease) were recruited to the study and outcomes related to these 12 participants are reported in Phase Two and Phase Three of the research. Of these 12 participants, 10 had a family member who participated in the intervention, described in Phase Three. The small sample size was justified given the in-depth characterisation and evaluation of participants, to examine the theoretical and clinical understanding of these clinical groups and their response to intervention. Where appropriate, data from individual participants are also explored to highlight variability between individuals and within dementia syndromes. Three distinct research phases are reported.

Phase One (Study 1) examined the evidence base for existing primary progressive aphasia and Alzheimer's disease lexical retrieval interventions, mapping each study to a theoretical framework of change mechanisms to identify whether particular mechanisms of change are associated with more effective outcomes, increasing understanding of potential mechanisms of change in progressive conditions and how these have, and may, be exploited in intervention. Additionally, this study aimed to investigate the role of nonlinguistic cognitive functions in the lexical retrieval studies reviewed.

Phase Two (Study 2 and 3) explored cognitive elements and discourse assessment in order to provide insights into diagnosis and assessment, with clinical implications for use of cognitive scaffolds in intervention and discourse as an intervention outcome measure. In the second study, sentence repetition deficits and evidence of error patterns using an adapted error classification schema were investigated, along with working memory abilities in which correlations are described. In the third study, stability of connected speech over successive sampling is examined to progress assessment tools to enable generalisation, beyond the word level, to be measured.

Phase Three (Study 4 and 5) evaluated a strategic self-cueing intervention, with a focus on generalisation outcomes, and the perceived experiences of both participants with dementia and their family members. In the fourth study, treatment effects for lexical retrieval performance (nouns, verbs, and adjectives), and generalisation effects for both untreated items and connected speech (communicative informativeness and efficiency) were investigated. The fifth study extended the evaluation of the self-cueing intervention through exploration of the perspectives and experiences of participants and their family members following participation in

intervention via thematic analysis of interview transcripts. Benefits of intervention beyond language gains and issues faced by these client populations and their families are explored.

The five studies, presented within the above three phases of research, provide evidence to support and inform future research with the impetus of developing meaningful, evidence-based speech pathology practice for lexical difficulties in dementia that is centered on optimisation of generalisation and is guided by the perspectives, insights, and needs of the client populations and their families.

Acknowledgements

This program of research would not have been possible without the research and clinical expertise of Associate Professor Anne Whitworth and Dr Jade Cartwright. You have both inspired and enabled me to pursue this research with an open and curious mind. Anne, your endless support, which knows no boundaries of time or place, has allowed a constant source of knowledge, guidance, and reassurance. Jade, you exude passion for best-practice services and pathways for people with neurodegenerative disease, and radiate an infectious pride for the speech-language pathology profession. You have played a quintessential role in my interest and confidence as both a clinician and researcher. Anne and Jade, thank you for the many rich, inquisitive, and inspiring discussions throughout this journey – it has been an absolute joy and has instilled in me a lasting sense of pride to be a part of our profession which I will carry with me into future clinical and academic endeavours. Thank you also to Professor Peter K Panegyres and Dr Robert Kane, who both generously offered their knowledge and expertise which has permitted this research.

Thank you to my parents, Kaye and Darrell, who have enabled an abundance of opportunities to nurture my education. Dad, thank you for your never-failing belief in my ability to achieve anything I put my mind to, and for always being there during moments when I forget. Thank you Ty, for your support and encouragement, for sharing the challenges and celebrations, and for the many timely reminders to take a break in nature, with Kubo.

Lastly, thank you to the participants and family members who generously offered their time to make this research possible. Each of you so kindly welcomed me into your home, and more pertinently, into your journey of living with progressive language difficulties. This research program is dedicated to you, your inspirational strength and willingness.

Publications Included in the Thesis

Beales, A., Whitworth, A., & Cartwright, J. (2018). A review of lexical retrieval intervention in primary progressive aphasia and Alzheimer's disease: Mechanisms of change, generalisation, and cognition. *Aphasiology*, *32*(11), 1360-1387.[1]

Beales, A., Whitworth, A., Cartwright, J., Panegyres, P. K., & Kane, R. T. (2019). Profiling sentence repetition deficits in primary progressive aphasia and Alzheimer's disease: Error patterns and association with digit span. *Brain and Language*, *194*, 1-11.[2]

Beales, A., Whitworth, A., Cartwright, J., Panegyres, P. K., & Kane, R. T. (2018). Determining stability in connected speech in primary progressive aphasia and Alzheimer's disease. *International Journal of Speech-Language Pathology*, *20*(3), 361-370.[3]

Beales, A., Bates, K., Cartwright, J., & Whitworth, A. (2019). Lost for words: Perspectives and experiences of people with primary progressive aphasia and Alzheimer's disease and their families of participation in a lexical retrieval intervention. *International Journal of Speech-Language Pathology*, *21*(5), 483-492.[4]

Submitted and currently under Review

Beales, A., Whitworth, A., Cartwright, J., Panegyres, P. K., & Kane, R. T. (*submitted*). Making the right connections: Maximising lexical generalisation in progressive lexical impairments through self-cueing, cognition, and communication partners**.** *American Journal of Speech-Language Pathology.*

[1] This is an 'Accepted Manuscript' of an article published by Taylor & Francis Group in Aphasiology, on 11 Jul 2018, available online: https://www.tandfonline.com/doi/full/10.1080/02687038.2018.1491192

[2] This is an 'Accepted Manuscript' of an article published by Elsevier in Brain and Language, on 3 July 2019, available online: https://www.sciencedirect.com/doi/full/10.1016/j.bandl.2019.03.001

[3] This is an 'Accepted Manuscript' of an article published by Taylor & Francis Group in International Journal of Speech-Language Pathology, on 8 March 2018, available online: https://www.tandfonline.com/doi/full/10.1080/17549507.2018.1442498

[4] This is an 'Accepted Manuscript' of an article published by Taylor & Francis Group in International Journal of Speech-Language Pathology, on 4 June 2019, available online: https://www.tandfonline.com/doi/full/10.1080/17549507.2019.1625439

Additional Publications Relevant to the Thesis

Beales, A., Cartwright, J., Whitworth, A., & Panegyres, P. K. (2016). Exploring generalisation processes following lexical retrieval intervention in primary progressive aphasia. *International Journal of Speech-Language Pathology*, *18*(3), 299-314.

Whitworth, A., Cartwright, J., **Beales, A.**, Leitão, S., Panegyres, P. K., & Kane, R. (2018). Taking words to a new level: A preliminary investigation of discourse intervention in primary progressive aphasia. *Aphasiology*, *32*(11), 1284-1309.

Authorship Attribution Statement

As lead author, I had primary responsibility for the conceptualisation and design of all studies. I completed the data collection, statistical analysis, and interpretation, taking the lead role in writing of all manuscripts. AW and JC contributed to conception and study design of all studies, and guided the method of recruitment and data collection. RK oversaw the statistical analysis conducted in three studies and provided statistical advice and support. KB assisted in the statistical analysis and interpretation in one study. PKP guided the recruitment process for the participants and oversaw interpretation of brain imaging data, along with provision of training in this area.

Funding: Dementia Australia Research Foundation and Curtin University through the Australian Government Research Training Program; Alzheimer's Australia Dementia Research Foundation [CDRN Half-Funded PhD Scholarship].

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Table of Contents

Chapter 1

Introduction

Chapter 1 presents the background and context for the research and the aims of the program of research. Clarification of key terminology implemented is provided. This chapter concludes with an overview of the thesis structure and methodology.

Dementia: A National Health Priority

Dementia is a growing global problem and has been identified as the greatest global challenge for health and social care in the 21st century, affecting 50 million people worldwide (World Health Organisation, 2017). In the United Kingdom, the proportion of people diagnosed with dementia has doubled from 2005 to 2015 (Donegan et al., 2017), with an estimated 850,000 people diagnosed with dementia in 2014 (Prince et al., 2014). In the United States, dementia prevalence estimates of 4 to 5 million have been reported (Plassman et al., 2007), with a growth up to three-fold predicted by 2050 (Prince et al., 2013). In Australia, dementia affects 10% of the Australian population over the age of 65 years and as many as 30% of those over 85 years of age (Australian Institute of Health and Welfare, 2016), with an estimated 459,000 Australians living with dementia in 2020 (Dementia Australia, 2018).

The clinical syndrome of dementia, recognised as a major neurocognitive disorder (DSM-5; American Psychiatric Association, 2013), describes the gradual decline in cognitive function that characterises more than 100 neurological disorders (Australian Institute of Health and Welfare, 2016; Sachdev et al., 2014). Alzheimer's disease (AD) is the most prevalent pathology and presentation of dementia cases (Australian Institute of Health and Welfare, 2012). Primary progressive aphasia (PPA) is considered a low prevalence syndrome of dementia (Mesulam et al., 2012). While precise estimates of the incidence and prevalence of PPA are not available (Grossman, 2010), the syndrome accounts for 20-40% of cases of frontotemporal dementia (Matias-Guiu & Garcia-Ramos, 2013). Individuals with progressive language disorders, associated with both PPA and AD, are less likely to be referred to speech-language pathologists than individuals with aphasia caused by stroke (Paul & Mehrhoff, 2015; Taylor et al., 2009). In a survey conducted by Code and Heron (2003), it was reported that speech-language pathologists in the United Kingdom

working with adults spent 3% of their time working with people with dementia, a concerning figure in consideration of the increased presentation of people with progressive language disorders such as PPA to clinical services (Taylor et al., 2009). With the prevalence of dementia increasing, irrespective of geographical demography (Prince et al., 2013), there is a pressing need to understand the nature of language difficulties in dementia and how these are best managed. Furthermore, gaining a greater understanding of the most appropriate mechanisms underpinning intervention and the key factors influencing intervention outcomes is critical in the global context of dementia care, ultimately to optimise quality of life.

Communication Difficulties Associated with Dementia

PPA and AD are two dementia syndromes that involve prominent communication difficulties (Gorno-Tempini et al., 2011; McKhann et al., 2011). Impaired language, or aphasia, is the foremost clinical feature of PPA, a group of heterogenous syndromes (Gorno-Tempini et al., 2011). While other cognitive functions, such as memory and visuospatial abilities, decline as the disease progresses, language is the main domain of dysfunction in at least the initial stages of disease (Mesulam, 2001). Language impairments in PPA are typically related to impaired word-level knowledge, including aspects of grammar, morphology, and functional category, and can impact reading comprehension and discourse and/or connected speech production (Thompson et al., 2013; Wilson et al., 2010). Impaired language functions also arise as a consequence of the AD pathophysiological process and can present early in the disease course (Blair et al., 2007; Forbes-McKay & Venneri, 2005), in which early features typically center on word finding or subtle conversational difficulties (Bayles et al., 1992). Impairments are also seen in verbal fluency (Cerhan et al., 2002; Henry et al., 2004), production of discourse and/or connected speech (Forbes et al., 2002; Forbes-McKay & Venneri, 2005), reading (Chapman et al., 2002; Croot et al., 1999), and writing (Croisile et al., 1996; Henderson et al., 1992).

Lexical retrieval difficulties, encountered when attempting to access words while speaking, are a common symptom of language impairment for both PPA (Mesulam, 2001; Rohrer et al., 2008) and AD (Bayles et al., 1992; Kavé & Goral, 2018). Lexical retrieval difficulties in PPA are associated with impairment of the language network and are a hallmark feature (Mesulam, 2001). Lexical retrieval

difficulties in AD are often reported to be subsequent to episodic or semantic memory loss (Bourgeois & Hickey, 2009), and/or impaired processing of sensory information (Kavé & Goral, 2018). The different manifestations of lexical impairment outlined here may result in breakdown at the macrolinguistic level, i.e. high order conceptual structures for organisation of information. Specifically, the informativeness of connected speech and/or discourse of people with svPPA are impacted by semantic errors, in addition to simplification of syntax and impaired local coherence (Boschi et al., 2017). The efficiency of connected speech of people with lvPPA are impacted by phonemic errors and reduced speech rate, which may be subsequent to impaired lexical retrieval (Wilson et al., 2010). Hallmark features of connected speech of people with AD include lexical errors, frequent use of high-frequency words, and hesitations (Boschi et al., 2017). To inform best-practice speech-language pathology interventions for people with PPA and AD, it is important to consider the underlying nature of impairment, with reference to theoretical models of lexical retrieval, as well as the impact that communication and lexical retrieval difficulties have on everyday conversations, social relationships, and ultimately, the individual's quality of life.

Classification of PPA Variants

PPA is a focal dementia that is characterised by the insidious onset and gradual progression of word finding or word comprehension impairments (Mesulam, 2001). Language impairment is seen in the context of relatively preserved episodic memory, visuospatial skills, reasoning, and social skills during the initial phase of the disease course. Nonlinguistic cognitive domains are typically affected after the first two years following onset, however, language remains the primary impaired function throughout the disease trajectory and is recognised to deteriorate faster than other domains (Mesulam, 2001). The language presentations of PPA coalesce broadly around three variants of the condition, a classification system that is underpinned by both the patterns of speech and language characteristics and the neuropathological presentations seen in this population. Current consensus criteria define the three variants of PPA as semantic variant (svPPA), logopenic variant (lvPPA), and nonfluent or agrammatic variant (nfvPPA), with classification of PPA into one of the variants occurring by clinical, imaging-supported, and/or definite pathologic diagnosis (Gorno-Tempini et al., 2011). Each variant presents with unique

speech and language features, which are central to clinical diagnosis, with lexical retrieval difficulties recognised as a core diagnostic feature of svPPA and lvPPA (Gorno-Tempini et al., 2011).

Using the current classification criteria, impaired confrontation naming and impaired single word comprehension are the core diagnostic features of svPPA (Gorno-Tempini et al., 2011). Three of the four secondary features must also be present, including impaired knowledge of low frequency and low familiarity items, surface dyslexia or dysgraphia, spared repetition, and/or spared grammar and motor speech (Gorno-Tempini et al., 2011). The imaging-supported classification of svPPA specifies evidence of damage involving predominant anterior temporal lobe atrophy and/or predominant anterior temporal hypoperfusion or hypometabolism on SPECT or PET (Gorno-Tempini et al., 2011). While lexical retrieval difficulties are present in other PPA variants, the disturbance in svPPA is considered most severe, particularly in comparison to other impacted language domains (Gorno-Tempini et al., 2011).

LvPPA is the most recently described variant of PPA (Gorno-Tempini et al., 2008) and, at present, is the least consistently defined variant, particularly in comparison to svPPA (Vandenberghe, 2016). Clinical classification of lvPPA includes two core features, impaired single word retrieval in spontaneous speech and naming, and impaired repetition of sentences and phrases (Gorno-Tempini et al., 2011). Three of the four secondary features must also be present, which include phonological errors in spontaneous speech and naming, spared single word comprehension and object knowledge, spared motor speech, and/or absence of frank agrammatism (Gorno-Tempini et al., 2011). The imaging-supported classification of lvPPA specifies evidence of damage in one of the following areas, predominant left posterior perisylvian or parietal atrophy on MRI, and/or predominant left posterior perisylvian or parietal hypoperfusion or hypometabolism on SPECT or PET (Gorno-Tempini et al., 2011). Notably, lvPPA is associated with substantially higher probability of the AD pathophysiological process (Leyton & Hodges, 2013). Since the publication of the Gorno-Tempini et al. (2011) classification criteria, it has been proposed that lvPPA encompasses two subtypes, one of which relates closely to linguistic AD while the other reflects the original diagnostic criteria provided by Gorno-Tempini and colleagues (Vandenberghe, 2016). Moreover, there is emerging research to support lvPPA as an atypical presentation of AD, evidenced by post

mortem findings of neurofibrillary tangles and amyloid plaques (Leyton et al., 2011; Mesulam et al., 2014), which has been reported to account for two-thirds of people diagnosed with lvPPA (Teichmann et al., 2013).

Clinical classification of nfvPPA features agrammatism in language production or apraxia of speech, that is, effortful, halting speech with inconsistent speech sound errors and distortions (Gorno-Tempini et al., 2011). Two of the three secondary features must also be present, including impaired comprehension of syntactically complex sentences, spared single word comprehension, and/or spared object knowledge (Gorno-Tempini et al., 2011). Notably, lexical retrieval difficulties are not a core diagnostic feature of nfvPPA. The imaging-supported classification of lvPPA specifies one of the following presentations, predominant left posterior fronto-insular atrophy on MRI, and/or predominant left posterior fronto-insular hypoperfusion or hypometabolism on SPECT or PET (Gorno-Tempini et al., 2011).

Classification of AD Presentations

AD dementia is a clinical syndrome that arises as a consequence of the AD pathophysiological process (Lindenboom & Weinstein, 2004). In the current clinical guidelines, criteria for diagnosis of AD requires two domains of cognitive or behavioural impairment, including impaired ability to acquire and remember new information, impaired reasoning and handling of complex tasks, impaired visuospatial abilities, impaired language functions, and/or changes in personality, behaviour, or comportment (McKhann et al., 2011). In the initial stages of AD, working memory is typically impaired, resulting in difficulties learning new information related to either semantic or episodic memory (Braaten et al., 2006). Language deficits in AD may present as difficulties in speaking, reading, and/or writing (Taler & Phillips 2008), characterised by lexical retrieval difficulties in the presence of impaired comprehension, particularly for abstract and complex information (Bourgeois & Hickey, 2009). Different presentations of AD are also recognised, the amnestic presentation and nonamnestic presentations, which present with different cognitive and language profiles (McKhann et al., 2011).

The amnestic presentation is the most common presentation of AD, in which memory impairment is the primary cognitive deficit (McKhann et al., 2011). Hallmark features include impaired learning and recall of recently acquired information, as well as impairment in at least one of the above outlined cognitive

domains. The neuroimaging in the amnestic presentation has been reported to show hippocampal atrophy (Mendez et al., 2012). In a study which examined the neuropsychological recognition of amnestic AD and lvPPA, performance on episodic memory, language, and visuospatial tasks were not found to significantly distinguish AD from lvPPA (Mendez et al., 2019). Notably, working memory was the only neuropsychological measure found to distinguish AD and lvPPA, which was found to be disproportionately decreased in lvPPA.

In the current clinical guidelines, three nonamnestic presentations of the pathophysiological process of AD have been defined, characterised by a dominant impairment in visuospatial, executive function, or language (McKhann et al., 2011). Impaired spatial cognition, including object agnosia, impaired face recognition, simultanagnosia, and alexia are the core features of the visuospatial presentation. In comparison to the amnestic presentation of AD, the neuroimaging in the visuospatial presentation has been reported to show more right parietal and occipital changes (Mendez et al., 2012). Hallmark features of the executive function presentation are impaired reasoning, judgment, and problem solving. The core feature of the language presentation is impaired lexical retrieval (McKhann et al., 2011). The neuroimaging in the language presentation, in comparison to the amnestic presentation, has been reported to show more left parietal changes (Mendez et al., 2012). Notably, this parietal atrophy seen in the language presentation of AD overlaps with the neuroimaging classification of lvPPA (Gorno-Tempini et al., 2011).

Impact of Communication Difficulties on Quality of Life

In a systematic review and meta-analysis, relationships and social engagement were identified as critical factors associated with reports of better quality of life for people living with dementia (Martyr et al., 2018). Communication difficulties experienced during everyday interactions can have profound implications for social connectedness in dementia (Clare et al., 2012; Pozzebon et al., 2016), which in turn may have adverse effects on quality of life. Communication difficulties evident in both PPA and AD, albeit with different etiologies and anticipated trajectories of progression, can have an overwhelming impact on conversation and the success of interaction. Consequently, speech-language pathology interventions designed to improve communication should have the dual aim of improving language abilities and making a meaningful difference to quality of life (Banerjee et

al., 2009; Worrall & Holland, 2003). Very few studies have explored quality of life in PPA, with only two empirical studies reported to date (Cartwright, 2015; Ruggero, 2017). Reduced communication confidence, social withdrawal and avoidance of speaking situations, and anxiety and distress related to language difficulties have been reported by people living with PPA, associated with relatively intact insight and awareness of language decline in the earlier stages of the disease course (Cartwright, 2015; Ruggero, 2017). Cartwright (2015) reported that people with PPA are at greater risk of poorer quality of life outcomes when access to timely diagnosis, intervention, and proactive support are restricted. Further research is, however, required to determine whether language interventions improve psychosocial outcomes across the variants of PPA. People with AD have been reported to experience increased frustration with loss of self-expression, reduced participation in social activities, and difficulty sustaining personal relationships (Potkins et al., 2003). In a qualitative study that involved people with early stage dementia, the importance of staying connected to others, associated with the diminishing ability to socially engage, was identified as a key theme in reducing feelings of loneliness (Moyle et al., 2011). Moreover, proxy-reported quality of life studies involving family members and caregivers have reported the importance of opportunity for engagement and social support as key indicators of quality of life for people with AD (e.g. Logsdon et al., 1999). Even in the advanced stages of AD, there is evidence of the significant emotional need for communication support (Astell & Ellis, 2006). Targeting communication to bolster social connectedness through accessible and systematic interventions, that have ecological validity to both the person with dementia and their family members, is critical to overcome the challenges of communication and reduce the impact on everyday life.

Family Member and Spousal Experience of Dementia

Spousal caregivers have reported disrupted communication as the greatest challenge, associated with increased negative perceptions of relationship quality and reduced connectedness (Braun et al., 2009; de Vugt et al., 2003; Pozzebon et al., 2016). Even early stages of dementia are associated with challenges across communication-related functions, highlighting the critical support role that spouses and family members can provide (Badarunisa et al., 2015; Woodward, 2013). As communication abilities decline with disease progression, the spousal relationship is

reported to become increasingly imbalanced and poses significant emotional load on the spouse (de Vugt et al., 2003; van Vliet et al., 2011). In order to adapt to communication changes and develop skills to support interaction, family members and spouses warrant early and ongoing professional guidance and proactive support, inclusive of speech-language pathology (Pozzebon et al., 2016; Woodward, 2013). The involvement of spouses and specifically, communication partner training, has been well established in the post-stroke aphasia literature (Simmons-Mackie et al., 2016) with some carry-over seen in the PPA literature (e.g. Grasso et al., 2017). In the AD literature, early implementation of communication interventions have been found to minimise the occurrence of changed behaviours which may have indirect benefits of supporting the individual to stay in their home environment due to improved communication within the spousal relationship (Arkin, 2007). Further development of communication interventions that support social interaction and maintenance of personal relationships for people living with dementia are needed (Woodward, 2013), which should routinely involve communication partners in strategy practice to target relevant contexts as well as enhance opportunities for strategy generalisation (Volkmer et al., 2019).

Speech-Language Pathology Interventions for Dementia

Despite the clear need for communication-focused supports for people living with dementia and their families, rehabilitative-style interventions (i.e. impairment-based) have not been routinely offered (Bourgeois et al., 2016; Code & Heron, 2003). Rather, speech-language pathology interventions have traditionally focused on indirect interventions, such as environmental adaptation, to support memory and communication difficulties (Royal College of Speech and Language Therapists, 2006). While speech-language pathologists receive specialist training in the assessment and treatment of communication disorders, people with PPA are under referred to speech-language pathology services in the United Kingdom (Volkmer et al., 2018) and Germany (Reidl et al., 2014), and it is unclear how many Australians with AD are referred to speech-language pathology services (Speech Pathology Australia, 2016). Moreover, an Australian study found that speech-language pathologist respondents reported lack of confidence in working with PPA (Taylor et al., 2009), while a study in the United States revealed that 43% of speech-language pathologists reported not being familiar with PPA (Wooley, 2014). The lack of

rehabilitative-style interventions, coupled with under referral of people with dementia to speech-language pathology services, highlights that "the traditional therapeutic nihilism that frequently accompanies diagnosis of progressive disorders" (Murray, 1998, p. 669) still prevails in practice, ultimately leading to the under recognition of the role of speech-language pathologists working with communication in dementia syndromes (Bourgeois et al., 2016). This is despite the growing evidence base for rehabilitative-style interventions in both PPA (Jokel et al., 2014) and AD (Woodward, 2013), as well as the shift in speech-language pathology practice statements which now highlight that interventions for people with dementia should be based on communicative strengths and weaknesses (Royal College of Speech and Language Therapists, 2014). Moreover, there has been advocacy from individuals living with dementia, affirming that speech-language pathologists are rarely included in the care plans of people with dementia despite individuals actively seeking support for word finding and other language difficulties (Swaffer, 2015).

To address the therapeutic nihilism and improve access to rehabilitative-style interventions, it is important to understand the current evidence base and directions for future research. Although previously deemed unsuitable for rehabilitation and impairment-based language and communication interventions due to the progressive neurodegeneration, increasing evidence to challenge this traditional view is being reported in both PPA (Carthery-Goulart et al., 2013) and AD (Morello et al., 2017). With lexical retrieval difficulties being prominent, the majority of the PPA intervention research aimed at directly targeting language impairments has focused almost exclusively on lexical retrieval intervention (Jokel et al., 2014). In studies of people with PPA, picture naming practice has been targeted through repeated naming with orthographic and phonological representations (e.g. Croot et al., 2015), spoken and/or written definitions (Jokel et al., 2010), phonological and/or semantic cues (e.g. Macoir et al., 2015), semantic feature analysis (e.g. Marcotte & Ansaldo, 2009), generative naming (e.g. Beeson et al., 2011), constraint-induced language therapy (e.g. Hameister et al., 2017), and self-cueing strategies (e.g. Beales et al., 2016). In contrast, there has been a lack of investigation of language and communication interventions for people with AD, with a predominant focus on memory-based approaches (Morello et al., 2017). Despite evidence of lexical retrieval difficulties in AD due to decline in the semantic system (Verma & Howard, 2012), cognitive rehabilitation approaches typically involve errorless learning (Haslam et al., 2010;

Metzler-Baddeley & Snowden, 2005), as well as a combination of mnemonic development, verbal elaboration, vanishing cues, and expanding rehearsal (e.g. Clare et al., 2002). Interestingly, in a systematic review of language and communication intervention in AD, Morello et al. (2017) found greater levels of evidence for approaches classified as lexical-semantic (i.e. those that involved semantic tasks during word learning), indicating promise of efficacy for this approach in AD. Uniquely, Flanagan et al. (2016) implemented semantic feature training for two participants with AD and found improved lexical retrieval for treated items post-intervention despite severe memory deficits, providing preliminary evidence to support language based interventions in AD.

Direct treatment gains following lexical retrieval interventions have been demonstrated in PPA through immediate positive gains, typically quantified through an increase in performance in spoken word naming measures post intervention (Carthery-Goulart et al., 2013). Although there is strong evidence to support direct treatment gains following memory rehabilitation approaches in AD through errorless learning, cue vanishing, and trial and error techniques (De Vreese et al., 2001), there has been limited investigation of gains related to language deficits (Woodward, 2013). Emerging evidence of greater levels of evidence following lexical-semantic approaches, albeit a small number of studies and with methodological limitations, highlight the need for application of language approaches in AD with robust and rigorous investigation (Morello et al., 2017).

In contrast to direct treatment effects, it is less clear how generalisable these gains are in terms of the transfer of treatment effects beyond therapy targets or contexts. Generalisation outcomes are integral to understanding how to maximise the meaningful benefits of intervention to everyday communication, and ultimately, make a positive difference to the quality of life of people living with dementia. Generalisation effects may be *within level*, i.e., change at the same linguistic level as targeted in therapy (Webster et al., 2015), also described as *horizontal* generalisation (Milman, 2016), and/or *across level*, i.e., change at a different linguistic level to the focus of therapy (Webster et al., 2015), also referred to as *vertical* generalisation (Milman, 2016). In a systematic review of generalisation and maintenance of treatment gains in PPA, individuals with svPPA were found to show limited within level generalisation following therapy as well as difficulty maintaining direct treatment gains (Cadório et al., 2017). By comparison, individuals with lvPPA and

nfvPPA were reported to have better capacity for within level generalisation to untreated items and maintenance. For example, in a picture naming with cueing hierarchy intervention involving one individual with lvPPA and one with svPPA, improved naming of untreated items was found for the lvPPA participant only (Newhart et al., 2009). Cadório et al. (2017) proposed that poor outcomes for people with svPPA might be due to impairment in semantic knowledge in which learning is rigid and requires practice in an individual's natural environment/context. For people with lvPPA and nfvPPA, Cadório et al. (2017) suggested that better scope for generalisation may be seen as lexical deficits are not item specific.

Alternatively, within level generalisation may be associated with lexical retrieval interventions that involve a strategy approach, as established in the post-stroke aphasia literature where generalisation of gains to untreated items has been more prevalent when a strategic approach has been used, i.e. once taught, a strategy can be applied to novel items (Nickels, 2002). For example, Henry et al. (2013) found generalisation to untreated items for both participants, one with svPPA and one with lvPPA, following a lexical retrieval self-cueing strategy. Moreover, Beales et al. (2016) also found within level generalisation for all participants, three with svPPA and one with lvPPA, following a self-cueing approach. Unique to the lexical retrieval studies reviewed by Cadório et al. (2017), Beales et al. (2016) included verb and adjective stimuli in addition to nouns, with direct treatment effects found for all participants and varying word class patterns for within level generalisation. In a systematic review of language and communication interventions in AD, generalisation outcomes were not reported (Morello et al., 2017). Similarly, there has been minimal investigation of generalisation outcomes following memory rehabilitation approaches in AD (De Vreese et al., 2001). Evaluation of across level generalisation (e.g. connected speech measures) has received minimal attention in both the PPA (Carthery-Goulart et al., 2013) and AD literature (De Vreese et al., 2001; Morello et al., 2017). Cadório et al. (2017) called for future studies to explore generalisation patterns that focus on discursive, i.e. discourse and conversation, outcome measures.

Despite the strong impetus for assessment and investigation of generalised gains beyond the word level following lexical retrieval intervention, limitations are evident in the current assessment tools for connected speech and discourse production. Challenges in discourse analysis include lack of standardised tasks and

measures as well as lack of investigation of the representativeness of connected speech measures over successive sampling, potentially impacting both accuracy and reliability of generalisation outcomes (Hird et al., 2006; Morello et al., 2017). Such limitations in the use of connected speech and discourse tasks extend to diagnosis, in which language assessment tasks are frequently based on single word or sentence processing despite established diagnostic features of connected speech in neurodegenerative diseases (Boschi et al., 2017). Although connected speech is recognised as a central component of language assessment in PPA and AD, there has been a lack of translation into clinical practice. Consequently, Boschi et al. (2017) called for further investigation of connected speech production in different neurodegenerative diseases to inform task and genre selection. Evaluation and development of assessment tools for utilisation in within and across level measurement to investigate generalised gains is warranted to better understand the theoretical underpinnings of intervention as well as the potential impact beyond the level targeted in treatment.

Theoretical Accounts of Lexical Retrieval in PPA and AD

Various psycholinguistic models have been applied to account for the phenomenon of lexical retrieval more generally, as well as the disruptions that may occur. These theoretical accounts of lexical retrieval have proposed both sequential and integrated frameworks.

Sequential Frameworks. The majority of these theoretical accounts are based on the classical Broca-Wernicke-Lichtheim-Geschwind lesion-deficit model of aphasia (Geschwind, 1965). This model underlies the classical paradigm of aphasia and assumes that there are brain centres and pathways dedicated to the storage and transmission of linguistic information (McNeil et al., 1991). A number of frameworks aim to understand lexical access as involving a sequential set of processes, where the lexical network is assumed to span three distinct levels of linguistic information; conceptual, semantic, and phonological (e.g. Blanken et al., 1987; Butterworth, 1989; Coltheart, 2004; Dell & O'Seaghdha, 1992). The conceptual system is proposed to occur prior to the lexical retrieval process, which results in the preverbal message (Blanken et al., 1987; Levelt, 1995; Rohrer et al., 2008). Impairment within this system may result in breakdown at the macrolinguistic level, i.e. the high order conceptual structures for organisation of information. In

connected speech and/or discourse, an overall reduced quantity of speech (Rohrer et al., 2008), reduced effectiveness in making references (Carlomagno et al., 2005), impaired thematic coherence (Glosser & Goodglass, 1990), and spontaneous echolalia or verbal stereotypies (Bathgate et al., 2001) are all features of AD, that may indicate a selective deficit at this level. Although not overtly addressed in sequential models, it has been suggested that macrolinguistic processing depends on multiple high order nonlinguistic cognitive processes, such as attention and memory systems, i.e. semantic, autobiographical, and episodic memory (Glosser & Goodglass, 1990). Moreover, there is speculation that executive functions underpin this conceptual stage of language processing and, consequently, may directly influence the nature and severity of impairment at this level (Carlomagno et al., 2005; Irwin et al., 2002; Rende et al., 2002; Rohrer et al., 2008). In contrast to AD, these executive processes are spared in early stages of PPA, although changes with disease progression in PPA are poorly understood (Etcheverry et al., 2012; Jefferies & Lambon Ralph, 2006).

The interactions or overlapping functions of these nonlinguistic factors should, therefore, be considered when evaluating language output, including connected speech and/or discourse production where they may be identified more readily. The output of the conceptual stage is the preverbal message, a concept that then activates the lexical-semantic representation within the semantic lexicon (Levelt, 1995). Rohrer et al. (2008) further refine this process into two components; word retrieval and verbal stores. Typically, deficits at this level will present in the form of semantic paraphasias, such as context-inappropriate word errors, e.g. ‘dog’ instead of ‘horse’, superordinate errors, e.g. ‘animal’ instead of ‘dog’, or circumlocutory errors, e.g. ‘they live in the garden and are brown’ (Jefferies & Lambon Ralph, 2006; Rohrer et al., 2008). Such errors are observed in AD (Lukatela et al., 1998) and svPPA (Rohrer et al., 2008). In AD, circumlocutions and semantic paraphasias may be interpreted as impaired semantic knowledge (i.e. verbal store), however, Rohrer et al. (2008) argue that the true nature of the deficit is in lexical retrieval. In contrast to svPPA, knowledge about words and the phonological encoding are preserved in AD; rather, it is the access to this information that is defective (Hillis, 2007). Consequently, single word comprehension is spared in early stage AD, while deficits are often reported in svPPA (Gorno-Tempini et al., 2004; Kirshner, 2012). Some cases of PPA may not reflect this pattern and lexical retrieval

difficulties may be observed in absence of impaired word comprehension, supportive of the heterogeneity that is characteristic of the primary progressive aphasias within and across variants (Vandenberghe, 2016). Moreover, the potential for an 'anomia only' variant of PPA has been suggested (Vandenberghe, 2016). Deficits of visual perception may also impact lexical retrieval through involvement of the verbal knowledge store (Rohrer et al., 2008). In comparison to early stage PPA, visuospatial functions are typically impaired in AD (Taler & Phillips, 2008). Such deficits may be observed on naming or description tasks, typically affecting low frequency items rather than common items (Rohrer et al., 2008). The semantic representation activates the lexical-phonological representation in the phonological output lexicon, a store that contains metrical information about the word, inclusive of the number of syllables, stress patterns, and segmental information (Levelt, 1995). Impaired access to the phonological representation often results in phonological paraphasias (Caramazza & Hillis, 1990; Howard & Gatehouse, 2006). The phonological structure may be altered through substitution e.g. 'crabon' for 'crayon', transposition e.g. 'aminal' for 'animal', omission e.g. 'elphant' for 'elephant', or addition processes e.g. 'hippopotoamus' for 'hippopotamus' (Rohrer et al., 2008). These errors are typically seen in lvPPA, associated with impaired phonological loop (Gorno-Tempini et al., 2008), one of the three components that comprise working memory (Baddeley, 1992). Further, Meyer et al. (2015) found that phonological short term memory was largely intact in early stage AD, while individuals with lvPPA demonstrated impairment on all tasks that required phonological short term memory. Within the working memory model proposed by Baddeley (1992), the phonological loop is responsible for maintaining a phonological representation of a novel word. Therefore, if the phonological loop was unable to store forms as correct or complete, lexical-semantic representations would be inaccurate or incomplete, thereby resulting in the storage of faulty phonological representations (Baddeley et al., 1998). Acheson et al. (2011) found that the brain regions responsible for the phonological encoding in language production are also responsible for short term retention of speech sounds in the working memory, supporting the interaction of these language and nonlinguistic cognitive functions.

Integrated Frameworks. Although discrete clinical profiles of lexical retrieval can be elucidated through sequential models, such accounts may

underestimate the complex nature of language processing and the heterogeneity that are often reported within syndrome groups (Cahana-Amitay & Albert, 2015). Integrative frameworks focus on how pathology disrupts connectivity within the language networks in the brain (Sonty et al., 2007). Functional integration does not contradict the principle of functional segregation, but rather complements it. Specifically, it is understood that there are individual elements that sustain distinct roles in information processing, however, interactions between networks form patterns of both segregation and integration (Sporns et al., 2004). Neuroimaging studies have demonstrated that during language processing tasks, not only are the classical language centers within the left perisylvian cortex activated, but also additional brain areas that are typically involved in nonlinguistic functions, such as perception and intention (Crosson et al., 2005; Pulvermuller et al., 2003). Such findings have led to the proposal that language processing is a dynamic system, in which linguistic and nonlinguistic cognitive functions transpire from shared and distributed neural networks (Blumstein & Amso, 2013; Cahana-Amitay & Albert, 2014). These functions of linguistic and nonlinguistic cognition in the brain are a result of neurochemical interactions, networks, and pathways, and therefore are dependent on connectivity as well as the underlying structure (Eickhoff et al., 2009). The language system then acts as a broadly distributed neural network rather than specific domains of processing (Blumstein & Amso, 2013), with language processing depending on widely distributed cortical and subcortical neural systems (Matsumoto et al., 2004). While areas of language processing such as Broca's area are still recognised, the primary focus is increasingly on the inter-regional connections that bind these functional cortical areas together (Sonty et al., 2007). As noted by Tseng (1996), while speech-language pathologists primarily focus on the speech and language product, understanding the underlying nonlinguistic process is critical for the dynamic observation of language performance and for the design of targeted interventions.

Therapeutic Potential and Neuroplasticity in PPA and AD

Rehabilitative-style interventions remain highly promising with respect to delaying and altering the trajectory of language decline in neurodegeneration. In the post-stroke aphasia literature, theories of the mechanisms of change, i.e. how the treatment ingredients result in the predicted outcomes (Whyte et al., 2014), are

largely centered on recovery and compensation (Code, 2001), as well as underlying principles of neuroplasticity (Kleim & Jones, 2008). For recovery mechanisms, also referred to as restitution, neural systems are restored through physiological processes (Rothi & Horner, 1983). In accordance with Stern (2013), neural compensation refers to the maintained or improved performance during a specified task via the recruitment of brain networks that are not activated when the brain is healthy, that is, those that are normally used in non-affected individuals. This mechanism emphasises the functional connectivity and interactions within brain networks, consistent with integrated and dynamic theories. Within the AD literature, there is evidence of prefrontal recruitment as a compensatory mechanism in response to functional loss. Studies have found that individuals with AD had increased activity in the prefrontal regions compared with healthy age-matched controls during cognitive tasks, interpreted as compensatory reallocation of cognitive resources (Backman et al., 1999; Becker et al., 1996; Grady et al., 1993). Grady et al. (2003) measured neural activity during semantic and episodic memory tasks in people with early stage AD and healthy elderly controls. For both groups, similar neural regions were activated in both tasks, including the prefrontal and occipital cortex. Different patterns of connectivity, however, were found in the two groups. Specifically, in both tasks, healthy controls recruited a left hemisphere network of regions, including the temporal and occipital cortex (Grady et al., 2003). Similarly, AD participants recruited a network that included the temporal and occipital cortex, but involved both hemispheres. Moreover, recruitment of this network of regions correlated with increased performance for AD participants, thus, providing evidence of the use of additional neural resources in the prefrontal cortex. This activity has been attributed to the mediation of executive functions in order to compensate for loss of cognitive function in semantic and episodic memory tasks (Grady et al., 2003). While there are potentially different mechanisms within the PPA and AD literature underpinning treatment effects, e.g. prophylactic versus restorative approaches, further investigation is required to better understand how the underpinning mechanisms produce the desired outcomes. Furthermore, future studies should consider and apply principles of neuroplasticity (Kleim & Jones, 2008) to enhance treatment and generalisation outcomes, applicable to both recovery and compensation approaches.

In the traumatic brain injury literature, application of internal memory strategies, i.e. behaviours or techniques designed to help a person gain control of

their learning and recall ability, have been well documented (O'Neil-Pirozzi et al., 2016,). Self-cueing strategies for lexical retrieval can be considered a form of internal memory strategy, requiring conscious thinking of ways to encode material. The growing evidence base for internal memory strategies has led to the recommendations from the Cognitive Rehabilitation Task Force that cognitive rehabilitation should target everyday function, include active attempts to promote generalisation, and involve direct application of compensatory strategies to functional contexts (Cicerone et al., 2019). Moreover, Sohlberg et al. (2005) reported the importance of instructional techniques, specifically the centrality to cognitive intervention for both restorative and compensatory techniques for persons with impaired memory and/or executive functions. Evidence to support the use of strategy-based instruction has been well documented within the learning disability literature (e.g. Swanson, 1999). Core instructional components of strategy-based instruction have been identified, inclusive of explicit practice, orientation to task, modeling of steps, and use of systematic probes for reinforcement (Swanson, 1999). Despite considerable interest in the neuroscience of compensation, particularly the impact of behaviour on brain activity, and the growing evidence in the traumatic brain injury and learning disability literature for use of metacognitive strategies, there is limited understanding of whether compensation of language impairments can be taught through use of strategies in dementia syndromes, potentially drawing on similar mechanisms.

Thesis Aims

This program of research comprises five unique lines of enquiry, presented within three interrelated phases of research. The aims of each research phase are outlined below.

In order to inform the development of a lexical retrieval intervention in Phase Three, specifically, how mechanisms of change can be exploited in intervention, Phase One examines the evidence base for existing PPA and AD studies, with specific aims to (1) identify which mechanisms of change have been applied to lexical retrieval intervention studies for people with PPA and AD, and whether particular mechanisms of change were associated with more effective outcomes, (2) determine whether particular mechanisms of change of lexical retrieval intervention

were associated with within and across level linguistic generalisation, and (3) identify the role of nonlinguistic cognitive functions in the reviewed studies.

Informed by the gaps in intervention design and interpretation found in Phase One, Phase Two explores two factors related to assessment. With implications for capacity for new learning and engagement in intervention, the primary aims of the first study were to (1) examine sentence repetition in people with PPA and AD, using an error classification schema adapted from Hohlbaum et al. (2018), and (2) explore correlations with digit span abilities, tasks known to draw on verbal working memory. Also informative to the planning and evaluation of the lexical retrieval intervention in Phase Three, the second study in Phase Two examines the viability of connected speech as an outcome measure, with specific implications for across level generalisation. The aims of this study were to (1) examine the stability of connected speech over three consecutive weeks in people diagnosed with PPA and AD on measures of lexical content, fluency, and communicative informativeness and efficiency, and (2) identify differences in stability of these measures in different discourse sampling tasks, specifically everyday monologues, narrative, and picture description.

Building on increased awareness of cognitive factors with potential implications for intervention and an increased confidence in using connected speech to monitor change, Phase Three of the research program evaluates a strategic self-cueing intervention with specific aims to (1) identify direct treatment effects for spoken word naming for treated items, (2) explore generalisation outcomes for spoken word naming of untreated items (within level generalisation), and (3) examine generalisation outcomes in connected speech (across level generalisation) as measured by communicative informativeness and efficiency. The second study in Phase Three extends the evaluation of the strategic self-cueing intervention using a qualitative methodology, with specific aims to (1) explore the perspectives and experiences of people with PPA and AD and their family members following an impairment-based lexical retrieval intervention, (2) explore any perceived benefits from participation in the intervention, and (3) inform future practice and service delivery models.

Research Methodology Overview

To address the above aims, participants were recruited to the study via private speech-language pathologists, neurologists, and geriatricians from rural, regional, and metropolitan areas of Western Australia. In total, 12 participants (four svPPA, four lvPPA, and four AD) were recruited to the study, aged between 55 and 86 years (M = 65.5) (see Table 1). All diagnoses were established by the referring neurologist or geriatrician via brain imaging and neuropsychological testing, and based on the current international consensus criteria for PPA (Gorno-Tempini et al., 2011) and diagnostic guidelines for AD (McKhann et al., 2011). A detailed analysis of language profiles and nonlinguistic cognitive functions was undertaken for all participants, with relevant details provided in the associated study.

A mixed methods research design incorporating quantitative and qualitative data (diagnostic group and individual) was utilised in this research program. The research methodology specific to each research question is reported in the associated study. Due to the timing of recruitment, data collection, and/or data analysis, participant data were not included in all studies. The third study addressing stability of connected speech was undertaken and published prior to all participants being recruited. Moreover, due to comprehension deficits, three participants (one svPPA and two AD) were unable to complete the interview methodology used in the qualitative exploration of participant experiences and their data was not included in the final published study.

This study was approved by the Curtin University Human Research Ethics Committee (HR218/2015).

Thesis Overview

The five interrelated studies are presented in five chapters (Chapters 2 to 6) (see Figure 1), preceded by the current Introduction (Chapter 1) which presents the background for the research, and followed by a Discussion (Chapter 7) which integrates the key research findings. Given the different lines of enquiry that comprise this work, each chapter is introduced before progressing to the next in order to highlight the relationship between the chapters and how each builds to achieve the overarching aim of the thesis. With the background and context for the research and the overall aims of the program of research being set out in Chapter 1, the following chapters set out the five studies.

Chapter 2, comprising one published study, presents the first phase of the research program, and examines the evidence base for existing PPA and AD lexical retrieval interventions, mapping each published study to a theoretical framework of change mechanisms to identify whether particular mechanisms of change are associated with more effective outcomes.

Chapter 3, comprising one published study, investigates the role of working memory in relation to diagnosis in PPA and AD, with specific focus on sentence repetition and digit span performance.

Chapter 4, comprising one published study, reports the stability of connected speech over successive sampling sessions which was analysed using measures of lexical content, fluency, and communicative informativeness and efficiency.

Chapter 5, comprising one study which is submitted and under review and therefore presented in chapter form, presents the intervention outcomes of a metacognitive strategy intervention for lexical retrieval, specifically, a strategic self-cueing approach with embedded nonlinguistic cognitive scaffolds. Diagnostic group level analyses and evaluation of individual data is reported.

Chapter 6, comprising one published study, presents the perspectives and experiences of participants and their family members following participation in the strategic self-cueing intervention through thematic analysis of interview transcripts.

Chapter 7 summarises and integrates the key research findings in light of the research objectives and their links with, and contribution to, the literature on lexical retrieval interventions in PPA and AD. In this final chapter, a summary of the major clinical implications is provided with suggestions for future research, and an outline of the key strengths and limitations of the research.

Table 1

Overview of participant demographics and corresponding participant numbers in Phase Two and Phase Three of the research program

	Participant											
Age (years)	62	55	72	72	60	70	64	59	86	60	68	58
Gender	M	M	M	M	F	M	F	F	M	F	M	M
Diagnosis	svPPA	svPPA	svPPA	svPPA	lvPPA	lvPPA	lvPPA	lvPPA	AD	AD	AD	AD
Education (years)	18	16	10	14	16	14	10	9	9	13	12	16
Time post diagnosis (years)	2	3	3	1	3	3	1	1	1	2	4	4
Phase Two*												
Study 2	1	2	3	4	5	6	7	8	9	10	11	12
Study 3	1	2	3	n/a	4	5	n/a	6	7	8	n/a	9
Phase Three*												
Study 4	1	2	3	4	5	6	7	8	9	10	11	12
Study 5	1	9[1]	2	n/a	3	4	8	5	6	7	10[1]	11[1]
Study 5 Family member	1	8	2	n/a	3	4	n/a[2]	5	6	7	9	10

* Numbers refer to allocated participant number in each corresponding paper. "n/a" indicates that participants' data were not included in the paper due to timing of recruitment, data collection, and/or data analysis. [1] Participant unable to participate in interview. [2] Family member did not participate in intervention or interview.

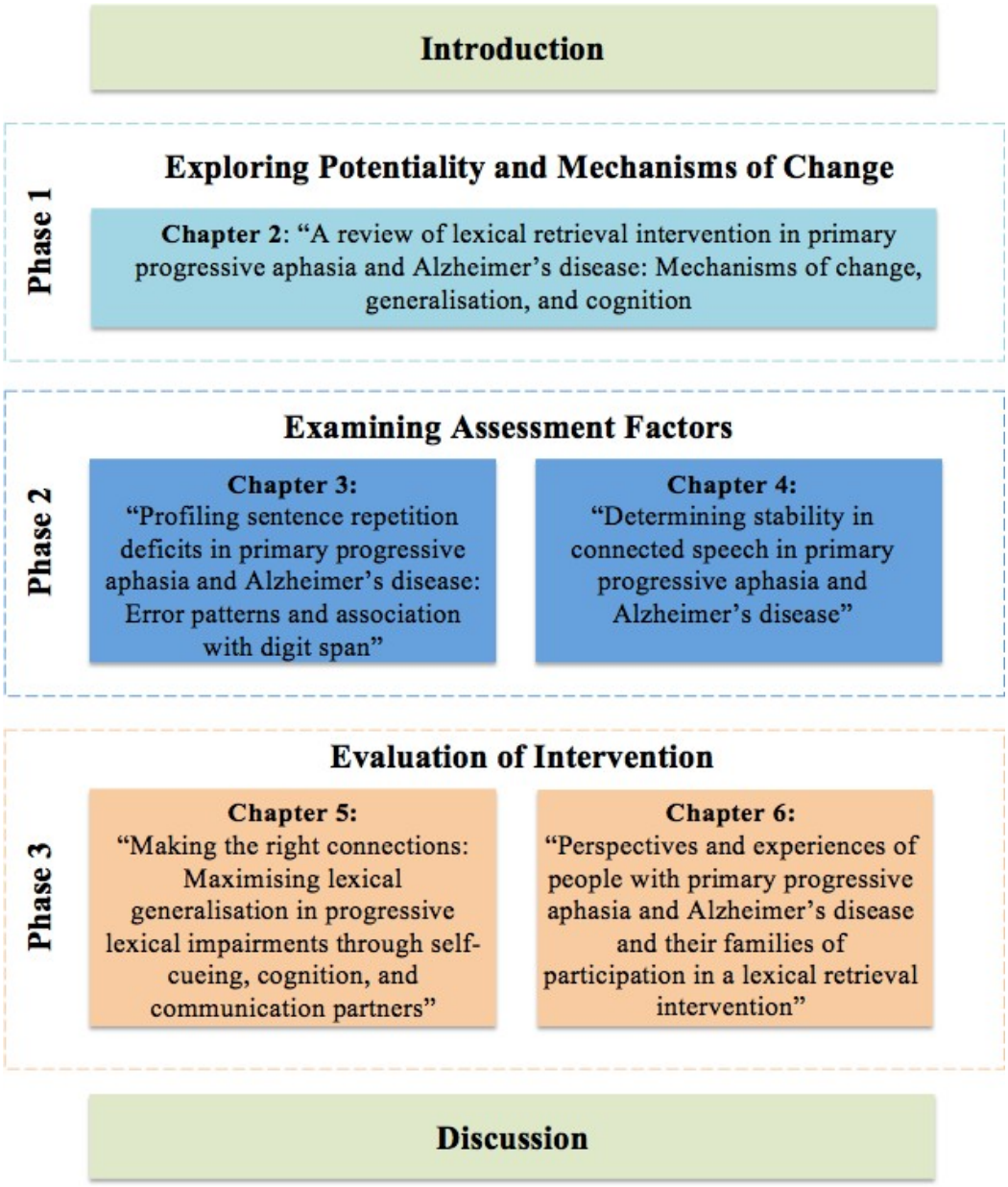

Figure 1

Thesis outline

PHASE ONE

Exploring Potentiality and Mechanisms of Change in Lexical Retrieval Interventions in PPA and AD

Chapter 2

Chapter 2 presents the findings of Phase One of the research that aimed to examine the evidence base for existing PPA and AD lexical retrieval interventions and map each study to a theoretical framework of change mechanisms to identify whether particular mechanisms of change are associated with more effective outcomes.

Study Overview

Building on the background information provided in Chapter 1, this study reviewed the evidence base for existing lexical retrieval treatment studies that have been applied in the PPA and AD literature. This review included all rehabilitative-style intervention approaches that targeted improved lexical retrieval, inclusive of designs that drew on linguistic processes (e.g. semantic memory) and/or nonlinguistic cognitive processes (e.g. episodic memory). As stated in Chapter 1, theories of mechanisms of change have been proposed in the post-stroke aphasia literature, and are often associated with recovery or compensation (Code, 2001). To understand the theoretical underpinnings of intervention approaches undertaken to date and identify opportunities to direct the field and future intervention research, studies were mapped using accounts of change mechanisms offered within the post-stroke aphasia literature. Recovery and compensatory change mechanisms, adapted for lexical retrieval, were drawn from theoretical frameworks of motor recovery (Code, 2001; Levin et al., 2009). Specifically, two mechanisms of lexical recovery; stimulation and relearning, and two mechanisms of lexical compensation; reorganisation and cognitive-relay, are applied. Additionally, the role of nonlinguistic cognitive functions in lexical retrieval interventions was investigated, inclusive of executive functions, attention, working memory, autobiographical memory, and episodic memory. Through evaluation of the PPA and AD intervention literature from a unique and critical perspective, findings of change mechanisms associated with the greatest promise for treatment outcomes are used to inform intervention planning and interpretation in Phase Three.

Beales, A., Whitworth, A., & Cartwright, J. (2018). A review of lexical retrieval intervention in primary progressive aphasia and Alzheimer's disease: Mechanisms of change, generalisation, and cognition. *Aphasiology*, *32*(11), 1360-1387.

Aphasiology

ISSN: 0268-7038 (Print) 1464-5041 (Online) Journal homepage: https://www.tandfonline.com/loi/paph20

A review of lexical retrieval intervention in primary progressive aphasia and Alzheimer's disease: mechanisms of change, generalisation, and cognition

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To cite this article: Ashleigh Beales, Anne Whitworth & Jade Cartwright (2018) A review of lexical retrieval intervention in primary progressive aphasia and Alzheimer's disease: mechanisms of change, generalisation, and cognition, Aphasiology, 32:11, 1360-1387, DOI: 10.1080/02687038.2018.1491192

To link to this article: https://doi.org/10.1080/02687038.2018.1491192

APHASIOLOGY
2018, VOL. 32, NO. 11, 1360–1387
https://doi.org/10.1080/02687038.2018.1491192

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REVIEW

A review of lexical retrieval intervention in primary progressive aphasia and Alzheimer's disease: mechanisms of change, generalisation, and cognition

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ABSTRACT

Background: While significant benefits of lexical retrieval intervention are evident within the primary progressive aphasia (PPA) and Alzheimer's disease (AD) literature, an understanding of the mechanisms that underlie change is limited. Change mechanisms have been explored in the post-stroke aphasia literature and offer insight into how change occurs through interventions with progressive language disorders. Exploration of change mechanisms may progress our understanding as to how and why generalisation is likely, or not, to occur, as well as gain insight into the non-linguistic cognitive functions that may play a role.

Aims: This review of the literature aimed to (1) map the mechanisms of change that have been proposed or hypothesised within the PPA and AD lexical retrieval intervention literature to a theoretical framework based on a framework of motor recovery following stroke and accounts of change mechanisms within the post-stroke aphasia literature and explore whether particular mechanisms of change were associated with more effective outcomes; (2) determine whether particular mechanisms of change were associated with within- and across-level linguistic generalisation, and (3) investigate the role of non-linguistic cognitive functions in the lexical retrieval intervention studies reviewed here.

Main Contribution: A search of Medline, PsycINFO, and CINAHL identified 37 papers published between 1982 and April 2016 that reported lexical retrieval intervention in people with PPA or AD, categorised here according to whether the proposed change mechanism was stimulation (12 studies), relearning (21 studies), reorganisation (three studies), or cognitive-relay (two studies). Significant treatment gains, predominantly based on linguistic performance measures, were reported for both diagnostic groups in association with the proposed mechanisms of stimulation and relearning. Significant treatment gains were also reported for people with PPA in association with reorganisation and cognitive-relay mechanisms; these mechanisms were only employed in PPA studies. Varying outcomes for linguistic generalisation were reported in 26 PPA and six AD studies. Nineteen studies incorporated non-linguistic cognitive functions in intervention; these were limited to autobiographical memory (17 studies), episodic memory (three studies), or both (one study).

ARTICLE HISTORY
Received 25 July 2017
Accepted 24 April 2018

KEYWORDS
Primary progressive aphasia; Alzheimer's disease; lexical retrieval; mechanisms of change; generalisation; cognitive functions

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Supplemental data for this article can be accessed here.

Conclusion: This review highlights that individuals with PPA and AD benefit from lexical retrieval intervention, irrespective of the mechanism of change, and that linguistic generalisation was reported in studies proposing different change mechanisms. Insufficient exploration of the role of non-linguistic cognitive functions was highlighted with respect to assessment, planning intervention, and interpreting intervention outcomes. Recommendations are made, with a view to heightening our ability to interpret intervention outcomes.

Introduction

Lexical retrieval difficulties manifest across a range of aetiologies, including a number of neurodegenerative diseases. Similar to post-stroke aphasia, different presentations of lexical difficulties are seen across dementia syndromes, for example primary progressive aphasia (PPA), where lexical retrieval difficulties are a hallmark feature (Mesulam, 2001), and Alzheimer's disease (AD), where lexical retrieval difficulties are often reported to be subsequent to memory loss (Bourgeois & Hickey, 2009). Current consensus criteria for PPA identify three variants: semantic (svPPA), logopenic (lvPPA), and non-fluent variant (nfvPPA) (Gorno-Tempini et al., 2011). Each variant presents with unique speech and language features, which are central to differential diagnosis within this focal dementia. SvPPA is characterised by lexical retrieval difficulties, comprehension, and object recognition deficits (Ash et al., 2013); lvPPA features lexical retrieval difficulties and impaired repetition (Gorno-Tempini et al., 2011) as well as halting and disruptions (Ash et al., 2013); and deficits in nfvPPA include effortful speech, agrammatism, and, in some cases, speech apraxia (Gorno-Tempini et al., 2011). Notably, lexical retrieval difficulties are not a core diagnostic feature of nfvPPA. Different clinical presentations of AD are recognised within the literature, including the amnestic presentation and the left hemisphere-dominant type with prominent linguistic difficulties (McKhann et al., 2011). The language profile seen in both AD clinical presentations is frequently characterised by lexical retrieval difficulties in the presence of impaired comprehension, particularly for abstract and complex information (Bourgeois & Hickey, 2009).

Despite the degenerative nature of these syndromes, a growing body of evidence is testimony to the effectiveness of lexical retrieval interventions. Immediate treatment effects, which are, in many cases, maintained over periods ranging from one week to six months, indicate that improvement is possible (De Vreese, Neri, Fioravanti, Belloi, & Zanetti, 2001; Jokel, Graham, Rochon, & Leonard, 2014), suggestive of increased storage and access to lost or degraded lexical items. Disparity in generalisation outcomes, that is, the transfer of treatment effects beyond therapy targets, has been reported in the PPA (see Carthery-Goulart et al., 2013) and AD literatures (De Vreese et al., 2001). With the intervention approaches endorsed in the post-stroke aphasia literature being largely applied in the remediation of progressive lexical retrieval difficulties, it could be assumed that the same proposed mechanisms of change are at play, despite the different aetiologies that lead to distinctive linguistic and non-linguistic cognitive profiles across the diagnostic groups. The role of non-linguistic cognitive functions with respect to planning and interpreting intervention outcomes is unclear. With the growing

evidence base for successful interventions, particularly in PPA, the opportunity is present to take stock and explore whether the same mechanisms of change are hypothesised to be involved or whether alternative or additional treatment strategies and models are needed to work with lexical difficulties in progressive conditions.

Theories of mechanisms of change

In the PPA and AD literature, the theoretical mechanisms of change producing the predicted intervention outcomes have not been well defined. In accordance with Whyte et al. (2014), the mechanism of change embodies how the active ingredients of an intervention produce the predicted outcomes. In the post-stroke aphasia literature, theories to explain change as a result of intervention have predominantly focused on recovery and compensation (e.g., Code, 2001; Lesser & Milroy, 1993; Rothi & Horner, 1983). Recovery, also defined as restitution, suggests that damaged neural systems are restored through physiological processes such as axonal regeneration. Through recovery, it is assumed that the original neural pathways resume activity (Code, 2001). In contrast, compensation suggests that impaired functions are taken over by previously uncommitted cortical areas (Code, 2001; Rothi & Horner, 1983). More specifically, deliberate compensation for impaired functions is achieved through involvement of those that are spared.

Levin, Kleim, and Wolf (2009) proposed a framework that drew on the World Health Organisation International Classification of Functioning (ICF) (2001) model, specifically the impairment and activity levels, to explain how motor recovery takes place following stroke. Depending on the treatment goals and associated measures, interventions are designed to target motor recovery or compensation. Change through recovery or compensation may impact the neural level (i.e., health condition), where gains reflect change in neural tissue (e.g., increased fMRI signal within a given cortical area). In addition, change may be seen at the performance level (i.e., body function and structure) through movement outcome or accomplishment (e.g., reappearance of premorbid movement patterns). Lastly, change at the functional level (i.e., activity) may be indicated through evaluations or scales that measure function (e.g., scale of movement quality rather than motor patterns and task accomplishment). Regardless of the level, each is targeted with the underlying objective to change the behaviour/s of an individual or prevent decline (Levin et al., 2009). For the purposes of this review, we have adapted this model to lexical recovery and compensation through the application of theoretical accounts of mechanisms of change within the post-stroke aphasia literature (see Table 1).

Lexical recovery

Based on the post-stroke aphasia literature, lexical recovery is proposed to occur through stimulation or relearning mechanisms (Rothi & Horner, 1983). In the case of lexical recovery, stimulation is the natural activation of lexical items by exposure without strategy. Similarly, relearning aims to retrain a network in the brain that has been previously effective through active engagement (Plaut, 1996). Such mechanisms of recovery aim to shift an individual's performance towards the "normal" or premorbid level, as well as prevent maladaptive behaviours (Lesser & Milroy, 1993). At the neural

Table 1. Framework of recovery and compensation of lexical retrieval within the WHO-ICF.

ICF Level	Motor Recovery	Lexical Recovery	Motor Compensation	Lexical Compensation
Health condition (neural)	*Restoring function in neural tissue that was initially lost after injury.*	Observed activation in brain areas previously inactivated due to disease processes, suggestive of: "stimulation"—natural activation of lexical items by exposure without strategy or "relearning"—retraining a network in the brain through active engagement.	*Neural tissue acquires a function that it did not have prior to injury.*	Observed activation in alternative brain areas not normally observed in lexical retrieval, indicative of "reorganisation".
Body functions/structure (performance)	*Restoring the ability to perform a movement in the same manner as it was performed before injury.*	Restoring lexical retrieval outcomes and related task accomplishments through "stimulation" or "relearning" (see definitions above).	*Performing an old movement in a new manner.*	Training an alternate step in lexical retrieval, indicative of "cognitive-relay".
Activity (functional)	*Successful task accomplishment using limbs or end effectors used by nondisabled individuals.*	Quality of interactions and related activities based on ability and/or perception through lexical recovery.	*Successful task accomplishment using alternate limbs or end effectors.*	Quality of interactions and related activities based on ability and/or perception through lexical compensation.

Adapted from Levin et al.'s (2009) definitions of Motor Recovery and Motor Compensation at three different levels.

level, recovery is seen by increased neural activity within a given cortical area associated with lexical retrieval. At the performance level, recovery is seen through the reappearance of premorbid lexical outcomes and related task accomplishments. Finally, at the functional level, quality of interactions and activities are changed based on ability and/or perception through lexical recovery.

Lexical compensation

Based on the post-stroke aphasia literature, lexical compensation is proposed to occur through reorganisation or cognitive-relay mechanisms (Lesser & Milroy, 1993; Rothi & Horner, 1983). Reorganisation may be observed at the neural level through activation in alternative brain areas not normally observed in lexical retrieval (Rothi & Horner, 1983). Lexical retrieval interventions that utilise cognitive-relay aim to bring about compensation at the performance level. By drawing upon intact cognitive functions, an alternative step in lexical retrieval may be trained to support the accomplishment of target behaviour/s and thereby discourage maladaptive behaviours and ineffective strategies (Lesser & Milroy, 1993). Lexical compensation through reorganisation or cognitive-relay may also produce change at the functional level (i.e., quality based on ability and/or perception) in order to meet communicative goals.

Generalisation effects

In addition to therapeutic outcomes, generalisation outcomes are integral both to understanding how to maximise the benefits of intervention and to subsequent treatment design. Generalisation effects may be *within level*, i.e., change at the same linguistic level as targeted in therapy, and/or *across level*, i.e., change at a different linguistic level to the focus of therapy (Webster, Whitworth, & Morris, 2015). Drawing on Webster et al.'s (2015) framework, generalisation is regarded here at a linguistic level only and not in terms on overall *impact* of intervention, e.g., increased quality of life. Evidence of naming untreated items would therefore be evidence of generalisation (within level), as would evidence of increased sentence structure following lexical intervention (across level) or use of treated words within connected speech (across level). Within the PPA literature, within-level generalisation outcomes are limited, with only few studies reporting an improvement of untrained stimuli (see Carthery-Goulart et al., 2013, for a review). Moreover, across-level generalisation, specifically connected speech measures, has received minimal attention, with those studies that have addressed this showing inconsistent outcomes (Carthery-Goulart et al., 2013). Similarly, there has been minimal investigation into generalisation outcomes following therapy for AD (De Vreese et al., 2001). Further investigation is warranted to better understand how the mechanisms of change, as well as the linguistic and non-linguistic cognitive functions, may influence both treatment and generalisation outcomes.

Non-linguistic cognitive functions

When profiling the strengths and weaknesses of people with PPA, the focus has been on language given their prominent lexical difficulties, whereas for individuals with AD, the focus has been on memory systems. While non-linguistic cognitive functions are relatively spared within the early stages of disease progression in PPA (Mesulam, 2001), impairments in non-linguistic cognitive functions have been reported across stages of disease progression

(Etcheverry et al., 2012). Despite this research, there has been limited language intervention literature that has conducted and reported assessment of relevant non-linguistic cognitive functions or planned investigation or interpretation of treatment outcomes in view of those functions. Interestingly, a relatively recent increase has been seen in the number of studies investigating the relationships between linguistic and non-linguistic cognitive functions in the post-stroke aphasia literature (e.g., Cahana-Amitay & Albert, 2015). Executive functions, attention, and aspects of memory, in particular, have been identified as key non-linguistic cognitive functions that interact with language networks, and consequently, may be predictive factors for intervention outcomes (Helm-Estabrooks, 2002). Given the impaired executive functions associated with frontal lobe degeneration in early-stage AD (Lafleche & Albert, 1995) and later-stage PPA (Mesulam, Grossman, Hillis, Kertesz, & Weintraub, 2003), it is highly plausible that these functions may be even more implicated in the language difficulties seen in progressive disorders and, consequently, should be considered in treatment planning and interpretation of outcomes. Depending on the demands of the setting in which a lexical item is being retrieved, irrespective of aetiology, the influence of non-linguistic cognitive functions may vary and thereby impact an individual's response (McNeil, Odell, & Tseng, 1991). In the post-stroke aphasia literature, alterations to non-linguistic cognitive functions have been found to impact performance including dual task versus single task (Murray, Holland, & Beeson, 1998), task complexity and language stimuli (Villard & Kiran, 2015), and additional distractions, shifting task demands, as well as the reliance on auditory attention without corresponding visual support (Villard & Kiran, 2015). The impacts of such non-linguistic cognitive functions have not yet been explored directly in relation to the dementia syndromes, and yet would seem to be critical in terms of further investigation.

Aim

This review sought to map the mechanisms of change (e.g., stimulation, relearning, reorganisation, and cognitive-relay) identified in published lexical retrieval interventions with people with PPA and people with AD to intervention outcomes, including evidence of generalisation, and to consider the role of non-linguistic cognitive functions. Based on adaptation of the theoretical framework offered by Levin and colleagues and accounts of mechanisms of change in the post-stroke aphasia literature, this review sought to

(1) identify which mechanisms of change have been applied to lexical retrieval intervention studies for people with PPA and AD, and whether particular mechanisms of change were associated with more effective outcomes;
(2) determine whether particular mechanisms of change of lexical retrieval intervention were associated with within- and across-level linguistic generalisation; and
(3) identify the role of non-linguistic cognitive functions in the reviewed studies.

Method

The framework of recovery and compensation of lexical retrieval, drawing on the ICF (see Table 1), was developed prior to this review in order to classify lexical retrieval intervention papers on the basis of the proposed mechanisms of change, and the three

levels set out in Levin et al.'s (2009) account of motor recovery and motor compensation were incorporated. We drew on definitions of mechanisms of change in the post-stroke aphasia literature to adapt the framework for classification of lexical retrieval studies, rather than motor function. The criteria specified for the inclusion of studies in this review consisted of: (a) publication between 1982 and April 2016, (b) inclusion of adults diagnosed with PPA or AD, and (c) reported data on lexical retrieval intervention outcomes. Search terms were chosen to focus on adults who had received a diagnosis of PPA or AD (PPA, semantic dementia, non-fluent progressive aphasia, fluent progressive aphasia, AD) and involved lexical retrieval intervention (lexical retrieval, word finding, language, anomia, intervention, rehabilitation, therapy, treatment). Combinations of these search terms were used for the electronic database search of Medline, PsycINFO, and CINAHL. The quality of studies was considered in order to aid interpretation, with methodological ratings applied based on the RoBiNT scale (Tate et al., 2013); however, no exclusion criteria were implemented (see Supplemental material A for RoBiNT methodology ratings of PPA and AD studies). Each study was scrutinised for: (a) participants (i.e., gender, chronological age, diagnosis, time from onset based on first instances of language difficulties, and prominent symptoms), (b) intervention design (i.e., aim, items, nature and modality of stimuli, interaction and type of facilitation, dosage, and timing), (c) measures (i.e., neural, performance, and functional), and (d) outcomes (i.e., direct-treatment outcomes of target items and tasks, linguistic generalisation outcomes, and maintenance outcomes) (see Supplementary material B for the template of individually reviewed papers). Whereas PPA variant was noted, AD clinical presentation was not, as this was not consistently reported in the papers that involved people with AD. Using the framework we developed, the mechanism of change was classified as stimulation, relearning, reorganisation, and/or cognitive-relay. Where no explicit discussion of a proposed mechanism of change was provided by the author/authors, the mechanism was inferred based on information provided about the treatment approach. Linguistic generalisation outcomes were coded for within- and across-level change, using Webster et al.'s (2015) framework. Lastly, all papers were coded for non-linguistic cognitive functions considered in therapy (i.e., autobiographical memory, episodic memory, attention, working memory, and executive functions).

Results

Thirty-seven studies met the criteria for review, 28 studies involving people with PPA and nine involving people with AD.

Mechanisms of change and direct-treatment outcomes

In addressing the first question as to which mechanisms of change have been proposed within lexical retrieval intervention studies for people with PPA or AD and whether there is a correspondence with treatment outcomes, the studies are presented under the change mechanisms of stimulation, relearning, reorganisation, and cognitive-relay approaches, and their outcomes are discussed.

Stimulation

Of the 37 studies reviewed, 12 studies proposed stimulation as the mechanism of change, 10 studies with people with PPA and two with people with AD (see Table 2), where therapy aimed to effect change through repeated production of the lexical item. The mechanism of change was explicitly discussed in two of the 10 studies (Jokel, Rochon, & Anderson, 2010; Savage, Ballard, Piguet, & Hodges, 2013; Savage, Piguet, & Hodges, 2014). For example, Jokel et al. (2010) proposed that they used a rich stimulation approach through repetitive practice of word-picture pairing, consistent with the classification of stimulation used in this review (see Supplemental material C for appraisal of individual PPA studies, and Supplemental material D for AD studies). The stimulation tasks typically involved repeated naming following exposure to a picture. In most studies, the corresponding label was provided for repetition in either orthographic form (e.g., Mayberry, Sage, Ehsan, & Ralph, 2011) or auditory form (e.g., Savage et al., 2013). All studies reported linguistic performance measures; Jokel et al. (2010) additionally reported functional outcomes based on a quality-of-life scale. No neural measures were reported.

Outcomes. Improvements in direct-treatment outcomes, based on linguistic performance measures, were reported in all 12 studies. Interestingly, Meyer, Snider, Eckmann, and Friedman (2015) compared phonological and orthographic cueing treatments and reported no significant improvements in oral naming accuracy for either therapy; however, they found significant improvements in written naming accuracy and naming to definition for the orthographic treatment. Savage et al. (2013) presented personally meaningful descriptions of treated items for two of the four participants in addition to the orthographic and audio forms. All participants showed significant improvements in naming accuracy, with large effect sizes being reported for the two participants who were presented with personally meaningful descriptions. For four individuals with AD, Metzler-Baddeley and Snowden (2005) compared errorless and errorful picture naming treatment for objects, famous people, and novel photographs and found significant improvements following both therapy conditions for familiar material only. For novel material, significant improvements were reported following the errorless approach only. Additionally, Senaha, Brucki, and Nitrini (2010) reported improvements in naming treated items; however, no statistical analysis was completed. Jokel et al. (2010) reported no significant functional effects based on the Quality of Communication Life Scale© (Paul et al., 1997).

Relearning

Twenty-one of the 37 studies proposed relearning as the mechanism of change, 13 studies with people with PPA and eight with people with AD (see Table 3), in which therapy aimed to retrain a network of lexical items through the semantic, phonological, and/or orthographic components of treated items and tasks. Of the eight AD studies, the mechanism of change was explicitly discussed in four studies (Clare, Wilson, Breen, & Hodges, 1999; Clare et al., 2000; Clare, Wilson, Carter, Roth, & Hodges, 2002; Ousset et al., 2002) and was addressed in all PPA studies except McNeil, Small, Masterson, and Fossett (1995) and Newhart et al. (2009). For example, Snowden and Neary (2002) proposed that reacquisition of semantic facts suggested a relearning mechanism of change (see Supplemental material C and D). The majority of the relearning treatment designs aimed to strengthen the links between intact semantic representations and the corresponding phonological representations of the treated items (e.g., Henry, Beeson, & Rapcsak, 2008).

Table 2. Studies targeting stimulation mechanism of change.

Investigator(s)	Participant(s)	Intervention	Direct-treatment outcomes		
			Neural	Performance	Functional
Croot et al. (2015)*	1 lvPPA 1 nfvPPA	RRIPP picture naming with orthographic and phonological representations provided using PowerPoint	–	Significant improvements naming treated items in picture naming	–
Haslam et al. (2010)*	22 AD	Comparison of trial and error naming approach with three learning approaches: errorless learning, vanishing cues, and errorless learning and vanishing cues combined	–	No significant improvements naming treated items in picture naming in comparison to errorless learning, vanishing cues, and errorless learning and vanishing cues approach	–
Heredia et al. (2009)*	1 svPPA	Picture naming with orthographic and phonological representations provided using PowerPoint	–	Significant improvements naming treated items in picture naming	–
Jokel et al. (2002), (2006)*	1 svPPA	Picture naming with orthographic representation and written definition provided	–	Significant improvements naming treated items in picture naming	–
Jokel et al. (2010)	1 svPPA	Picture naming using MossTalk Words® software with spoken definition and orthographic representation provided	–	Significant improvements naming treated items in picture naming	No improvement on Quality of Communication Life Scale©
Mayberry et al. (2011)*	2 svPPA	Picture naming with orthographic representation provided	–	Significant improvements naming treated items in picture naming	–
Metzler-Baddeley and Snowden (2005)*	4 AD	Picture naming with errorful approach or errorless approach (orthographic representation provided)	–	Significant improvements naming familiar treated items in picture naming (errorful and errorless approach) Significant improvements naming novel treated items in picture naming (errorless only)	–
Meyer et al. (2015)*	1 lvPPA	Computer-based picture naming with phonological or orthographic representations provided	–	No significant improvements in oral picture naming Significant improvements in written picture naming and naming to definition (orthographic treatment only)	–
Savage et al. (2013)	4 svPPA	Computer-based picture naming with orthographic representation and audio recording of spoken word provided (personally meaningful descriptions for two participants)	–	Significant improvements naming treated items in picture naming	–
Savage et al. (2014)	5 svPPA	Computer-based picture naming with orthographic representation and audio recording of spoken word provided	–	Significant improvements naming treated items in picture naming	–
Senaha et al. (2010)*	3 svPPA	Picture and written definition naming with orthographic representation provided	–	Improvements naming treated items in picture and definition naming	–

*Mechanism of change inferred

Table 3. Studies targeting relearning mechanism of change.

Investigator(s)	Participant(s)	Intervention	Direct-treatment outcomes		
			Neural	Performance	Functional
Bier et al. (2009)	1 svPPA	Picture naming with formal-semantic therapy, as well as comparison of spaced retrieval and simple repetition method	–	No significant improvements naming treated items in picture naming for all methods	–
Clare et al. (1999)	1 AD	Mnemonic development, verbal elaboration, vanishing cues, and expanding rehearsal strategies for errorless learning picture naming	–	Significant improvements naming treated items in picture naming (photographs of friends)	–
Clare et al. (2000)	6 AD	Individually tailored intervention, e.g., mnemonic development, verbal elaboration, vanishing cues, and expanding rehearsal strategies for errorless learning picture naming	–	Significant improvements naming treated items in picture naming for five participants (photographs of friends and celebrities)	–
Clare et al. (2002)	12 AD	Mnemonic development, verbal elaboration, vanishing cues, and expanding rehearsal strategies for errorless learning picture naming	–	Significant improvements naming treated items in picture naming for seven participants (photographs of friends and celebrities)	No improvement on quality of life questionnaire
Dewar et al. (2009)	1 svPPA	Picture naming with orthographic and semantic cues provided, with participant-generated mnemonic and vanishing cues paradigm	–	Significant improvements naming treated items in picture naming (photographs of celebrities)	–
Dunn and Clare (2007)*	10 AD	Comparison of four interventions: paired associate, vanishing cues, target selection, and forward cues	–	Significant improvements naming treated items (photographs of novel faces and celebrities) with no significant differences between intervention approaches	–
Graham, Patterson, Pratt, and Hodges (1999), (2001)	1 svPPA	Repetition of words using dictionary and self-generated material word exercises	–	Significant improvements in treated category verbal fluency task	–
Haslam et al. (2010)*	22 AD	Comparison of three learning approaches: errorless learning, vanishing cues, and errorless learning and vanishing cues combined, with trial and error	–	Significant improvements naming treated items (photographs of novel faces) for all learning approaches in comparison to trial and error	–
Henry et al. (2008)	2 PPA (variant unspecified)	Clinician-guided retrieval using semantic tasks, i.e., identification of subcategories and attributes	–	Significant improvements in treated category verbal fluency task for one participant	–
Jokel et al. (2009)	2 nfvPPA	Picture naming using MossTalk Words® software and presentation of cueing hierarchy (initial grapheme, whole word, repetition)	–	Significant improvements naming treated items in picture naming	–
Jokel and Anderson (2012)	7 svPPA	Comparison of four semantic and phonological cueing interventions: errorless passive, errorless active, errorful passive, and errorful active	–	Significant improvements naming treated items in picture naming (errorless approaches only, no differences in active or passive)	–

(*Continued*)

Table 3. (Continued).

Investigator(s)	Participant(s)	Intervention	Direct-treatment outcomes		
			Neural	Performance	Functional
Laffan, Metzler-Baddeley, Walker, and Jones (2010)*	20 AD	Comparison of three interventions: errorless learning, errorless learning and self-generated responses, and trial and error	–	Significant improvements naming treated items (photographs of celebrities) for errorless intervention approaches, but more significant for the combination intervention	–
Macoir et al. (2015)	1 svPPA	Semantic and phonological cueing intervention naming the corresponding verb in action videos	–	Significant improvements naming treated items in video naming (cued items only)	–
McNeil et al. (1995)*	1 PPA (variant unspecified)	Generation of antonyms and synonyms following oral presentation of lexical items with cueing hierarchy	–	Improvements in antonym and synonym generation for treated lexical item targets	–
Newhart et al. (2009)*	1 svPPA 1 lvPPA	Picture naming with cueing hierarchy (written naming, reading, search for written naming, auditory repetition)	–	Significant improvements naming treated items in picture naming	–
Noonan, Pryer, Jones, Burns, and Ralph (2012)*	8 AD	Comparison of errorless (reading and repetition) and errorful (phonemic and orthographic cueing) picture naming	–	Significant improvements naming treated items for both intervention approaches	–
Ousset et al. (2002)	8 AD	Definition naming from narrative context (episodic and semantic reinforcement)	–	Significant improvements in naming to definition for seven participants	–
Robinson et al. (2009)	2 svPPA	Object naming with phonological, orthographic, semantic, and personal definition cueing	–	Significant improvements in treated object naming for one participant	–
Snowden and Neary (2002)	2 svPPA	Orthographic, phonological, and personal definition cueing intervention	–	Significant improvements naming treated items in picture naming for one participant	–
Suárez-González et al. (2015)	1 svPPA	Comparison of conceptual enrichment therapy (autobiographical experiences) and naming therapy (orthographic representation and description provided)	–	Significant improvements naming treated items for both intervention approaches	–

*Mechanism of change inferred.

Outcomes. Similar to stimulation approaches, linguistic performance measures were the focus of direct-treatment outcomes. No studies incorporated neural measures, and only one study (Clare et al., 2002) included functional measures. Improvements based on the linguistic performance measures were reported for at least one participant in 20 of the studies. Interestingly, Haslam, Moss, and Hodder (2010) found that all learning approaches (i.e., errorless learning, vanishing cues, and errorless learning and vanishing cues combined) enhanced improvement of naming accuracy significantly in comparison to the trial and error approach. Unlike the other treatment designs, the trial and error approach was the only one that did not employ relearning; rather, stimulation was the inferred mechanism of change. The one study that did not report intervention gains (Bier et al., 2009) involved one participant with svPPA. Bier et al. (2009) proposed that the lack of direct-treatment gains, based on linguistic performance measures, might be associated with insufficient number of relearned semantic attributes required to restore the link between the participant's semantic and phonological representations.

Reorganisation

Three of the 37 studies proposed reorganisation as the mechanism of change (see Table 4), promoting alternative parts of the brain to take over the impaired function. All studies involved people with PPA and targeted activation of alternative brain areas as compensation for language loss. The intervention tasks consisted of generative naming and feature analysis (Beeson et al., 2011; Marcotte & Ansaldo, 2010) and repetition with cueing support (Dressel et al., 2010). All studies reported neural and linguistic performance measures, while Beeson et al. (2011) also reported functional measures.

Outcomes. Based on the comparison of fMRI data conducted during the linguistic performance task pre- and post-intervention, Beeson et al. (2011) reported increased activation of the left dorsolateral prefrontal regions. For one participant with nfvPPA, pre-intervention activations were bilateral, suggestive of gradual bilateral compensation (Marcotte & Ansaldo, 2010). Interestingly, larger post-intervention activation maps in semantic processing areas were found in comparison to pre-intervention ones. Specifically, temporal areas in the left hemisphere (superior temporal gyrus and middle temporal gyrus) were recruited when naming treated items following intervention. Similarly, Dressel et al. (2010) reported that fMRI data identified changes in cortical activity within the right superior and inferior temporal gyrus, suggesting to the authors that compensatory reorganisation mechanisms were occurring in the right hemisphere. Intervention gains, based on linguistic performance measures, were found in all studies; however, no statistical analysis was completed in the Marcotte and Ansaldo (2010) study. Beeson et al. (2011) reported functional gains based on post-treatment interview, specifically, the participant reported increased vocabulary and confidence in daily communication; this was not, however, analysed statistically.

Cognitive-relay

Of the 37 studies reviewed, two studies proposed cognitive-relay as the mechanism of change (see Table 5) whereby studies each trained a strategy to embed or encourage an alternative step in the lexical retrieval process, assuming lexical compensation rather than recovery. Both studies involved people with PPA, specifically, three people with svPPA and one person with lvPPA (Beales et al., 2016), and one person with svPPA and

Table 4. Studies targeting reorganisation mechanism of change.

			Direct-treatment outcomes		
Investigator(s)	Participant(s)	Intervention	Neural	Performance	Functional
Beeson et al. (2011)	1 lvPPA	Generative naming and feature analysis	fMRI showed increased activation of the left dorsolateral prefrontal regions	Significant improvements naming treated items in picture naming	Improved confidence and vocabulary on self-assessment questionnaire and interview
Dressel et al. (2010)	1 svPPA	Picture naming with phonological and semantic cues	fMRI showed changes in cortical activities: right superior and inferior temporal gyrus	Significant improvements naming treated items in picture naming	–
Marcotte and Ansaldo (2010)	1 nfvPPA	Semantic feature analysis	fMRI showed increased activation of temporal areas in the left hemisphere	Improvements naming treated object and action picture naming	–

Table 5. Studies targeting cognitive-relay mechanism of change.

			Direct-treatment outcomes		
Investigator(s)	Participant(s)	Intervention	Neural	Performance	Functional
Beales et al. (2016)	1 lvPPA 3 svPPA	Self-cueing strategy comprising semantic, phonological, orthographic, and autobiographical cues	–	Significant improvements naming treated items in picture (definition naming for participant four)	Improved confidence in word finding and strategy use on self-assessment questionnaire
Henry et al. (2013)	1 lvPPA 1 svPPA	Self-cueing hierarchy comprising semantic, phonological, orthographic, autobiographical cues and generative naming tasks	–	Significant improvements naming treated items in picture naming	Improved perception of communication abilities on self-assessment questionnaire

one person with lvPPA (Henry et al., 2013), and explicitly discussed the potential mechanism of change. Both authors proposed that spared cognitive functions were drawn upon in order to train a strategy and improve lexical retrieval (see Supplemental materials C and D). The strategy reported in each was a self-cueing hierarchy involving semantic, phonological, and orthographic cues, as well as autobiographical cues in the Beales et al. (2016) study. Both studies reported linguistic performance measures and functional measures, with neural measures not taken.

Outcomes. Significant intervention gains, based on the linguistic performance measures, were reported in both studies. Functional gains were reported, although not analysed statistically, on the basis of improved participant perception of communication abilities on self-assessment questionnaires, specifically improved accuracy and confidence in retrieving words (Beales et al., 2016) and overall improved confidence in communication (Henry et al., 2013).

Linguistic generalisation outcomes

To address the second question as to whether evidence of within- and across-linguistic level generalisation corresponded with the proposed mechanism/s of change, studies are presented within generalisation-level categories and outcomes discussed with respect to generalisation of the linguistic behaviours within or across levels. Word-level generalisation is primarily captured in discussion of within-level change, while generalisations to sentence and discourse levels are captured in discussions of across-level changes.

Within level

All studies, with the exception of Dunn and Clare (2007), Haslam et al. (2010), Metzler-Baddeley and Snowden (2005), and Snowden and Neary (2002) who reported no linguistic generalisation measures, reported on generalisation outcomes for word-level effects (see Table 6). Of the stimulation studies, generalised word effects were reported in 10 of the 12 studies, reporting the following outcomes involving people with PPA; improved exemplar naming, i.e., naming a different exemplar to the one treated (Croot

Table 6. Studies reporting generalisation outcomes.

			Generalisation outcomes	
Mechanism of change	Investigator(s)	Participant(s)	Within level	Across level
Stimulation	Croot et al., 2015	1 lvPPA 1 nfvPPA	Improved naming of exemplar pictures of treated items	No improvement in retrieval of target items Improved words overall, content words, and closed class words
	Heredia et al. (2009)	1 svPPA	No improvements in naming untreated items	Anecdotal from caregiver (increased use of treated items in daily communication)
	Jokel et al. (2002), (2006)	1 svPPA	No improvements in naming untreated items	–
	Jokel et al. (2010)	1 svPPA	No improvements in naming untreated items	No syntactic improvements on Oral Sentence Production Test
	Mayberry et al. (2011)	2 svPPA	Improved naming of exemplar pictures of treated items (one participant)	–
	Meyer et al. (2015)	1 lvPPA	Significant cross-language improvement in oral naming in Norwegian (orthographic therapy only)	–
	Savage et al. (2013)	4 svPPA	No improvements in naming untreated items	–
	Savage et al. (2014)	5 svPPA	No improvements in naming untreated items Improved comprehension of treated (not untreated) items in home context	Improved word retrieval of trained items (in video descriptions) for four participants
	Senaha et al. (2010)	3 svPPA	No improvements in naming untreated items	–

(*Continued*)

Table 6. (Continued).

Mechanism of change	Investigator(s)	Participant(s)	Generalisation outcomes	
			Within level	Across level
Relearning	Bier et al. (2009)	1 svPPA	No improvements in generating verbal attributes	–
	Clare et al. (1999)	1 AD	Improvements in naming in real-life setting for treated photographs	–
	Clare et al. (2000)	6 AD	Improvements in naming in real-life setting for treated photographs for two participants	–
	Clare et al. (2002)	12 AD	No improvements in naming untreated items	–
	Dewar et al. (2009)	1 svPPA	Improved naming of exemplar pictures of treated items	–
	Graham et al. (1999), (2001)	1 svPPA	No improvements in verbal fluency of untreated categories	–
	Henry et al. (2008)	2 PPA (variant unspecified)	No improvements in verbal fluency of untreated categories	–
	Jokel et al. (2009)	2 nfvPPA	No improvements in naming untreated items	Syntactic improvements on Oral Sentence Production Test
	Jokel and Anderson (2012)	7 svPPA	Improved naming of untreated items	No syntactic improvements on Oral Sentence Production Test
	Laffan et al., 2010	20 AD	No improvements in naming untreated items	
	Macoir et al. (2015)	1 svPPA	No improvements in naming untreated items	–
	McNeil et al. (1995)	1 PPA (variant unspecified)	Improved retrieval of untreated items	No improvement in words per minute or CIU per minute
	Newhart et al. (2009)	1 svPPA 1 lvPPA	Improved naming of untreated items in treated category (lvPPA participant)	–
	Noonan et al. (2012)	8 AD	No improvements in naming untreated items	
	Ousset et al. (2002)	8 AD	No improvements in untreated naming to definition items	
	Robinson et al. (2009)	2 svPPA	Improved naming of untreated objects (1 participant)	Improved definitions of objects
	Suárez-González et al. (2015)	1 svPPA	No improvements in naming untreated items	Improved description of treated items (conceptual enrichment therapy only)
Reorganisation	Beeson et al. (2011)	1 lvPPA	Improved naming of untreated items	Improved CIU per minute and speaking rate
	Dressel et al. (2010)	1 svPPA	No improvements in naming untreated items	–
	Marcotte and Ansaldo (2010)	1 nfvPPA	No improvements in naming untreated items	–
Cognitive-relay	Beales et al. (2016)	1 lvPPA 3 svPPA	Improved naming of untreated items	Improved CIU per minute and percentage for all svPPA
	Henry et al. (2013)	1 lvPPA 1 svPPA	Improved naming of untreated items	–

Table 7. Non-linguistic cognitive functions incorporated in intervention.

			Cognitive functions	
Mechanism of change	Investigator(s)	Participant(s)	Autobiographical memory	Episodic memory
Stimulation	Croot et al. (2015)	1 lvPPA 1 nfvPPA	Selection of treated items based on personally relevant topics	–
	Jokel et al. (2002), (2006)	1 svPPA	Treated item definitions derived from participant	–
	Savage et al. (2013)	4 svPPA	Therapy included personally meaningful definitions (two participants only) and photographs of participant's household items as treated items	–
	Savage et al. (2014)	5 svPPA	Treated items consisted of photographs of participant's household items	–
Relearning	Bier et al. (2009)	1 svPPA	–	Use of spaced retrieval principles
	Clare et al. (1999)	1 AD	Treated items consisted of photographs of friends	–
	Clare et al. (2000)	6 AD	Treated items consisted of photographs of friends and celebrities	–
	Clare et al. (2002)	12 AD	Treated items consisted of photographs of friends and celebrities	Use of spaced retrieval principles
	Dunn and Clare (2007)	10 AD	Treated items consisted of photographs of celebrities	–
	Graham et al. (1999), (2001)	1 svPPA	Therapy included self-generated material	–
	Laffan et al. (2010)	20 AD	Therapy included self-generated responses and treated items included photographs of celebrities	–
	Ousset et al. (2002)	8 AD	–	Therapy included episodic reinforcement
	Robinson et al. (2009)	2 svPPA	Therapy included personal definition cueing and treated items included personal household objects	–
	Snowden and Neary (2002)	2 svPPA	Therapy included personal definition cueing	–
	Suárez-González et al. (2015)	1 svPPA	Therapy included autobiographical experiences	–
Cognitive-relay	Beales et al. (2016)	1 lvPPA 4 svPPA	Therapy included autobiographical cues	–
	Henry et al. (2013)	1 lvPPA 1 svPPA	Therapy included autobiographical cues	–

et al., 2015; Mayberry et al., 2011) and improved naming in an untrained language (Meyer et al., 2015). Interestingly, Savage et al. (2014) reported improved comprehension of treated items in the context of the participant's home (i.e., verbal requests provided by family member); however, no improvements were found for untreated items. The remainder of the studies reported no significant improvements in naming untreated items (Croot et al., 2015; Heredia, Sage, Ralph, & Berthier, 2009; Jokel et al., 2010; Jokel, Rochon, & Leonard, 2002, 2006; Savage et al., 2013, 2014; Senaha et al., 2010). Significant word effects were reported in seven of the 21 relearning studies, specifically, improved retrieval of untrained items was found for people with PPA (Jokel & Anderson, 2012), whereas Newhart et al. (2009) reported improvements for individuals with lvPPA but not for individuals with svPPA, and Robinson, Druks, Hodges, and Garrard (2009) reported improvement for one of the two individuals with svPPA. Improved retrieval of untreated items was reported by McNeil et al. (1995); however, this was not supported by statistical analysis. In addition, improved exemplar naming for one person with svPPA (Dewar, Patterson, Wilson, & Graham, 2009) and improved naming of trained items in real-life setting (e.g., Clare et al., 1999) were found. Of the reorganisation studies, Beeson et al. (2011) reported word-level generalisation through improved naming of untreated items. Both cognitive-relay studies found improved naming of untreated items following intervention (Beales, Cartwright, Whitworth, & Panegyres, 2016; Henry et al., 2013).

Across level

Although the reviewed studies targeted lexical retrieval in isolation, six studies investigated generalised sentence effects (Jokel, Cupit, Rochon, & Leonard, 2009; Jokel et al., 2010; Jokel & Anderson, 2012; Robinson et al., 2009; Savage et al., 2014; and Suárez-González et al., 2015) (see Table 6). Three relearning studies found generalised sentence effects, specifically, Jokel et al. (2009) found improved syntax on the Oral Sentence Production Test (OSPT) for two people with nfvPPA, Suárez-González et al. (2015) found improved descriptions of treated items for one person with svPPA (following conceptual enrichment therapy only), and similarly, Robinson et al. (2009) reported improved sentence definitions of treated and untreated items, as rated by the assessor on a three-point scale (i.e., poor = 0; adequate = 1; or good = 2), for two people with svPPA. Savage et al. (2014), in a stimulation study, found improved word retrieval of trained items (in video descriptions) for four people with svPPA. Lastly, Jokel et al. (2010), a second stimulation study, and Jokel and Anderson (2012), a relearning study, also used the OSPT; however, they found no syntactic improvements for people with svPPA.

In addition to sentence effects, five studies investigated generalisation at the discourse level (Beales et al., 2016; Beeson et al., 2011; Croot et al., 2015; Heredia et al., 2009; and McNeil et al., 1995) (Table 6). Beales et al. (2016), a cognitive-relay study, reported improved measures of correct information units (CIU) (Nicholas & Brookshire, 1993) for three people with svPPA in nine monologic everyday discourse samples. In addition, Beales and colleagues found improvements in word class counts, with word class patterns varying across individual participants. Similarly, Beeson et al. (2011), in a reorganisation study with people with PPA, found improved CIU per minute and speaking rate in a picture description task (the "birthday picture" from Nicholas & Brookshire, 1993). Croot et al. (2015) found no improvements in the retrieval of target items; however, they reported improved content and closed class words for one lvPPA and

one nfvPPA participant. McNeil et al. (1995) also found no improvement in words or CIU per minute for one person with PPA. Lastly, for one individual with svPPA, Heredia et al. (2009) reported increased use of treated items in daily communication, as provided as anecdotal evidence from the caregiver.

Non-linguistic cognitive functions

The final question sought to identify which non-linguistic cognitive functions have been incorporated into therapy to date and whether they have been considered influential to the outcome. Inclusion of the non-linguistic cognitive functions, specifically, autobiographical memory, episodic memory, attention, working memory, and executive functions, were reviewed across the studies. Autobiographical memory, comprising episodes from the individual's life, has been found, at both neural and functional levels, to differ from episodic memory more generally and they are regarded here as separate stores (see Gilboa, 2004, for discussion). Nineteen studies were found to incorporate non-linguistic cognitive functions in intervention; these were limited to autobiographical memory (17 studies), episodic memory (three studies), or consideration of both (one study) (see Table 7). These comprised five stimulation studies, 12 that proposed relearning, and the two cognitive-relay studies. No studies aimed at reorganisation explicitly incorporated non-linguistic cognitive functions in therapy. Autobiographical memory was incorporated into therapy through the use of personally relevant treatment items (e.g., Clare et al., 1999), topics (e.g., Croot et al., 2015), definitions (e.g., Jokel et al., 2002, 2006), and cueing prompts (e.g., Beales et al., 2016). Episodic memory was incorporated into therapy using spaced retrieval principles (Bier et al., 2009; Clare et al., 2002), a technique that requires individuals to rehearse information at different and increasing spaced intervals of time. Furthermore, episodic reinforcement was targeted through the prior exposure of treated items in a narrative task (Ousset et al., 2002). Despite the assessment of a range of non-linguistic cognitive functions in the initial profiling of participants (e.g., working memory, attention, visuospatial; see Supplemental materials C and D for assessment details), these factors were not considered in therapy.

Discussion

In this review, we have adapted and applied a mechanism of change framework comprising stimulation, relearning, reorganisation, and cognitive-relay, organised within the broader categories of restitution and compensation, to characterise the direct-treatment gains in the PPA and AD lexical retrieval intervention literature. We have examined the within- and across-level linguistic generalisation outcomes with respect to Webster et al.'s (2015) framework and identified which non-linguistic cognitive functions have been considered in therapy.

Mechanisms of change and direct-treatment outcomes

Overall, relearning was the most prominent mechanism of change within the PPA and AD literature, employed in 21 of the 37 studies reviewed. The frequency of this change mechanism was followed by stimulation (12/37), while reorganisation (3/37) and cognitive-relay (2/37) were observed less. Across the four mechanisms of change reviewed, significant treatment gains were reported for each of the different proposed mechanisms.

The majority of studies reported improvements based on performance measures for at least one participant, with the exception of Bier et al. (2009), who reported no gains following treatment, and Meyer et al. (2015), who found no significant improvements in oral naming accuracy. It is important to note that although Marcotte and Ansaldo (2010), McNeil et al. (1995), and Senaha et al. (2010 reported improvements in naming accuracy, no statistical analysis was completed. Given the reporting of significant treatment gains following intervention, across each of the change mechanisms, there is no evidence yet to suggest that better treatment outcomes are associated more with one type of mechanism compared to another. Moreover, significant treatment gains were found for both client populations. Interestingly, reorganisation and cognitive-relay intervention designs were only employed in PPA; investigation of these change mechanisms for people with AD is warranted. These findings do, however, highlight that, as in post-stroke aphasia, change can be facilitated through each of these mechanisms.

Mechanisms of change and generalisation

In addition to direct-treatment outcomes, this review set out to explore whether evidence of within- and across-level generalisation corresponded with particular change mechanisms. While the majority of studies in the review included measures for linguistic generalisation at the word level, a number of studies also assessed for across-level generalisation effects, specifically, the sentence and discourse levels. With regard to within-level generalisation, improvements were reported in four of the stimulation studies, seven relearning studies, one reorganisation study, and both of the cognitive-relay studies. The remaining studies that assessed within-level generalisation but found no improvement consisted of five stimulation studies, 11 relearning studies, and two reorganisation studies. Less than half of the stimulation, relearning, and reorganisation studies, therefore, resulted in within-level generalisation (i.e., 14 of the 33 studies). While the cognitive-relay studies were too few in number to draw any strong conclusions, evidence of change in both was promising. Of the 11 studies that reported across-level generalisation outcomes, gains were reported for two stimulation studies, three relearning studies, one reorganisation, and one cognitive-relay study, while two stimulation and two relearning studies reported no improvement. Again, with significant gains reported for within- and across-level generalisation across the different groups of studies, it is clear that no one mechanism of change was more likely than another to facilitate generalisation following intervention. The lack of prediction of generalisation outcomes based on the mechanism of change alone leads us to a more multifaceted explanation of what might cause change beyond the direct target of a given intervention. Further investigation into the active ingredients of therapy is an area of enquiry that may also reveal factors that enhance generalisation outcomes. Such factors may include treatment ingredients (e.g., treatment tasks and/or materials, duration of treatment and/or frequency) and participant characteristics (e.g., diagnosis, time post onset).

Non-linguistic cognitive functions

Finally, in this review, we investigated the role of non-linguistic cognitive functions in lexical retrieval interventions for people with PPA and AD. Despite searching for a range of non-linguistic cognitive functions, including autobiographical memory, episodic

memory, attention, working memory, and executive functions, only autobiographical memory and episodic memory were incorporated into therapy designs.

While autobiographical memory was the most common non-linguistic cognitive function to be incorporated into intervention, this was seen in only 17 studies. Interestingly, 55.5% of the AD studies (5/9) incorporated autobiographical memory compared to 42.9% of the PPA studies (12/28). Typically, the incorporation of this memory component was to optimise personal relevance and importance in order to enhance treatment outcomes, consistent with the neural plasticity principle of salience (Kleim & Jones, 2008). For example, Clare et al. (1999) and Clare et al. (2000) used personal photographs of people as target items in treatment. In the PPA literature, Robinson et al. (2009) incorporated personal household objects to support naming. Similarly, Savage et al. (2013) explicitly targeted autobiographical memory through the use of photographs of personal household items as treatment items and personally meaningful definitions (with two of the four participants only). Savage et al. (2013) proposed that the personal descriptions were not a necessary component in the success of improved naming or maintenance as all four participants showed improvement; however, large effect sizes were reported for the two participants who received the personal descriptions. In addition, this study only included words in which participants demonstrated residual semantic knowledge. As suggested by Savage et al. (2013), a more enriched practice involving autobiographical items may be important when the treatment goal is to relearn concepts when no residual semantic knowledge is evident or suspected, rather than a more generalised stimulation approach. Croot et al. (2015) selected topics and stimuli based on informal conversation with the participant and their spouse prior to intervention (e.g., family and personal history, hobbies, and activities). In addition to optimising personal relevance and importance, autobiographical memory factors have been intentionally incorporated due to the preserved nature of autobiographical information in PPA. For example, to capitalise on spared memory systems, Beales et al. (2016) elicited autobiographical memories and used these to cue participants. Similarly, Suárez-González et al. (2015) drew upon autobiographical experiences to create a meaningful semantic environment to support access to target lexical items. This element of self-generation has been proposed to support better retention of information, in comparison to passively received information (Slamecka & Graf, 1978). Although salience is already a key consideration in the treatment of aphasia, further investigation into how to best exploit this principle with progressive disorders is clearly warranted.

Despite semantic memory being pivotal to the majority of studies through direct activation of the semantic lexicon, Bier et al. (2009) and Macoir et al. (2015) proposed that intervention gains might be a result of improved access to episodic memory, rather than restored semantic networks. For example, Henry et al. (2008) found that a participant with impaired episodic memory based on pretreatment cognitive profiling made minimal gains on linguistic performance measures, in comparison to a participant with intact episodic memory who demonstrated significant improvements. In the initial stages of disease progression, episodic memory is assumed to be intact in PPA (Gorno-Tempini et al., 2011), yet only one PPA study incorporated episodic memory into the intervention design. Interestingly, the person with svPPA in Bier et al.'s (2009) study, in which no significant treatment outcomes were found, was reported to have working memory and inhibition deficits determined on pretreatment profiling. Although these deficits were identified, the potential impact of the non-linguistic cognitive functions was not

considered in treatment design or interpretation of outcomes. In the AD literature, Clare et al. (2002) also used spaced retrieval principles, while Ousset et al. (2002) included episodic reinforcement through the prior exposure of treatment items in a narrative. Clare et al. (2002) and Ousset et al. (2002) found significant improvement for targeted items; however, despite incorporating episodic memory into intervention design, no significant improvements in linguistic generalisation outcomes were reported. It is important to note that theoretical discussions about memory systems, as well as the treatment methods attributed, remain inconclusive. For example, in this review, spaced retrieval has been attributed to episodic memory, as per author reports; however, alternative memory systems, such as procedural memory (Mimura & Komatsu, 2007), may also be associated, consequently limiting the understanding drawn from this attributed method.

Embedding mechanisms of change in a theory of intervention

Attempting to apply a mechanism of change framework to a diverse range of studies involving different aetiologies and treatment approaches is bound to come up against limitations created by the equally diverse use of nomenclature. This review was no exception. Seminal work by authors such as Code (2001), Lesser and Milroy (1993), and Rothi and Horner (1983) around characterising mechanisms of change provides a strong foundation for understanding how interventions may effect change. In future research, attempts to map these frameworks to each other and agree on common terminology would be useful with respect to both understanding and comparing studies. This extends to gaining consensus around what constitutes "recovery", as well as the nuances between, for example, restitution and restoration, where a common nomenclature would be more likely to facilitate systematic reflection of these concepts when designing therapies.

For the majority of studies, the mechanism underlying change was explicitly discussed (23/37). The assignment of the hypothesised mechanism of change to the framework in this review revealed inconsistencies within the intervention literature. Specifically, some papers predicted change mechanisms based on the treatment approach, whilst others based it on the outcome measure. For example, studies that reported reorganisation could be classified as stimulation or relearning using the framework; however, the authors of these papers proposed that the mechanism of change was reorganisation, supported by brain imaging outcomes. Consequently, studies reporting reorganisation mechanisms have developed their predictions based on an outcome measure, rather than the theory of intervention. In this case, we have classified the change mechanism based on the author's reports; however, we acknowledge that there may be additional mechanisms at play. One exception in this review is the classification of the mechanism of change in Mayberry et al.'s (2011) study. Although the authors discussed relearning as the potential mechanism of change, through application of the framework, we classified stimulation as the change mechanism based on the treatment approach.

Code (2001) raised the issue of determining whether restitution or compensation mechanisms have taken place, even in studies in which the treatment is explicitly described. Kolk (2000) suggested that there may be different forms of compensation, distinguished by the degree of consciousness involved. Specifically, unconscious compensation occurs within a neural or cognitive system. In contrast, conscious attempts by an individual, and potentially the assistance of the therapist or conversation partner,

involves implementation of a strategy to compensate for lost function. In relation to this review, unconscious compensation may be indicative of reorganisation, while conscious compensation may reflect cognitive-relay.

Furthermore, we acknowledge that mechanisms of change may co-occur and, although not discussed to any extent in this review, investigation is warranted. For example, the majority of treatment designs involved an element of stimulation, with some combined with an additional mechanism such as relearning. Furthermore, other studies in this review may well have had an element of reorganisation, although neural imaging was limited to three studies, which is necessary to determine this. The potential for combined and interacting mechanisms of change further endorses the need for explicit treatment designs with clear hypotheses regarding the underlying cause of change associated with the therapy approach.

Limitations found from the evaluation of the methodological quality of the reviewed studies highlight implications for understanding the mechanisms of change, as well as the role of non-linguistic cognitive functions. For example, when reviewing the "baseline characteristics" (see Supplemental material A), although most studies reviewed (36/37) provided information regarding the demographic, medical, and functional status, or profile of test scores, only six studies articulated how these variables informed intervention. The consideration and evaluation of baseline characteristics prior to commencing intervention require authors to take into account theories about the roles of linguistic and non-linguistic cognitive functions, critical for understanding the mechanisms of change. Stricter adherence by authors, where possible, to methodological rating scales, such as the ROBinT (Tate et al., 2013), in the design and development of a theory of intervention will increase both the reliability and interpretability of findings. Consequently, this will impact our understanding of the mechanisms of change and other factors at play in intervention, including the role of non-linguistic cognitive functions.

By applying a framework of recovery and compensation of lexical retrieval to hypothesise mechanisms of change following intervention, this review has drawn attention to the need to continue to develop a theory of intervention in which change mechanisms are firmly embedded (Byng & Black, 1995; Whyte et al., 2014). As has been shown, 14/37 studies have not made clear predictions regarding the mechanisms of change, resulting in a lack of attention to those intervention ingredients that may play an active role in change. Application of a framework to predict change mechanisms will encourage detailed identification of critical aspects of therapy as well as any aspects that may be altered without impacting the underlying change mechanism. Hypothesising mechanisms of change calls for a precise means of defining, specifying, and measuring intervention, which will heighten our ability to interpret intervention outcomes.

Conclusion

In conclusion, significant potential for benefit from lexical retrieval intervention is evident for both PPA (all variants) and AD diagnostic groups, irrespective of the mechanism of change. To date, relearning has been the most prominent mechanism of change within the PPA and AD literature, while reorganisation and cognitive-relay change mechanisms have only been proposed within the PPA literature. Significant linguistic generalisation outcomes have been reported in studies with these two diagnostic groups, within and

across levels, although generalisation has not been systematically measured. Close parallels can be seen in the ways in which people with progressive conditions respond to treatments that have been borrowed from the post-stroke literature, reinforcing that existing approaches can be drawn on and that change can be expected in the former, less-investigated clinical populations. Further parallels are highlighted with the post-stroke literature in the need to focus on and develop our understanding of the wider issues around therapy. Finally, despite searching for a range of non-linguistic cognitive functions, only autobiographical memory and episodic memory have been incorporated into PPA and AD lexical retrieval intervention within the period of the review, highlighting that insufficient exploration of non-linguistic cognitive functions has taken place. The importance for clinicians and researchers to systematically profile non-linguistic cognitive functions in progressive conditions, with a view to both deepening our understanding of the conditions and inform subsequent intervention, is equally highlighted.

Disclosure statement

The authors report no conflicts of interest.

Funding

This work was supported by the Dementia Australia Research Foundation and Curtin University through the Australian Government Research Training Program; Alzheimer's Australia Dementia Research Foundation [CDRN Half-Funded PhD Scholarship]

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PHASE TWO

Examining Assessment Factors of Nonlinguistic Cognitive Functions and Measurement of Generalisation

Chapter 3

Chapter 3 presents the findings of the first study in Phase Two of the research that aimed to investigate working memory, with specific focus on sentence repetition and digit span, in relation to diagnosis in PPA and AD.

Study Overview

This study explored working memory, a critical nonlinguistic cognitive function identified in Phase One of this research program to have not been considered in lexical retrieval intervention design or interpretation. As outlined in Chapter 1, working memory deficits, which typically present in the early stages of AD, are associated with difficulties in new learning (Braaten et al., 2006). Furthermore, impairment of the phonological loop, a component of working memory, has received increasing investigation in lvPPA (e.g. Meyer et al., 2015). Although there has been increased attention to this cognitive function in lvPPA, the significance as a diagnostic criterion is poorly understood (Mesulam & Weintraub, 2014). In the post-stroke aphasia literature, the impact of working memory impairment on language intervention outcomes has been reported, specifically, greater response to intervention for individuals with less impaired working memory (e.g. Fillingham et al., 2006). Consequently, knowledge of deficits associated with dementia pathophysiological processes is critical in the assessment and planning of a language intervention given the known impact of underlying nonlinguistic cognitive functions on intervention outcomes (McNeil et al., 1991; Villard & Kiran, 2015). Despite integrative theoretical accounts of language (Sonty et al., 2007), the correlation between performance on verbal sentence repetition and verbal working memory tasks for people with progressive language impairments is not well understood. An understanding of the interaction between working memory capacity, as measured by digit span performance, and sentence repetition, may inform integrative theoretical accounts of language. To deepen our understanding of such associations, sentence repetition deficits and evidence of error patterns using an adapted error classification schema are examined, along with digit span abilities in which correlations are described. The potential impact of working memory on capacity for new learning, strategy uptake, and engagement in intervention for people living with PPA and AD is crucial to informing the intervention planning in Phase Three, as well as the interpretation of intervention outcomes.

Beales, A., Whitworth, A., Cartwright, J., Panegyres, P. K., & Kane, R. T. (2019). Profiling sentence repetition deficits in primary progressive aphasia and Alzheimer's disease: Error patterns and association with digit span. *Brain and Language*, *194*, 1-11.

Brain and Language 194 (2019) 1–11

Contents lists available at ScienceDirect

Brain and Language

journal homepage: www.elsevier.com/locate/b&l

Profiling sentence repetition deficits in primary progressive aphasia and Alzheimer's disease: Error patterns and association with digit span

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ARTICLE INFO

Keywords:
Primary progressive aphasia
Alzheimer's disease. sentence repetition
Working memory
Logopenic variant
Semantic dementia

ABSTRACT

The use of sentence repetition tasks to distinguish dementia syndromes, particularly variants of primary progressive aphasia (PPA), is receiving growing attention. Impaired sentence repetition is a core feature of logopenic variant PPA, although the underlying cognitive mechanisms of this impairment and its significance as a diagnostic criterion remain poorly understood. Sentence repetition abilities of 12 people with dementia, using an adapted error classification schema, were analyzed, along with digit span abilities, a measure frequently used to assess working memory capacity, to explore error patterns and correlations. Participants with semantic variant PPA showed the greatest performance on sentence repetition and digit span in comparison to the logopenic variant PPA and Alzheimer's disease participants. Sentence repetition errors were characterized by middle omissions for semantic variant PPA, ending omissions and phonological errors for logopenic variant PPA, and ending omissions and unrelated word substitutions for Alzheimer's disease. Significant correlations were found between sentence repetition and digit span abilities. Findings are discussed in relation to working memory capacity theories, specifically, the dual-component model.

1. Introduction

Primary progressive aphasia (PPA) and Alzheimer's disease (AD) are dementia syndromes that are part of a continuum of clinical and pathological manifestations (Gorno-Tempini et al., 2011; McKhann et al., 2011). Accurate etiological diagnosis is critical for patient management, however, has proven difficult given the heterogenic nature of these syndromes (e.g. Dickerson et al., 2017). The use of sentence repetition tasks to distinguish dementia syndromes, particularly variants of PPA, is receiving growing attention regarding its reliability and current recommendation as a diagnostic criterion (e.g. Mesulam & Weintraub, 2014).

PPA comprises a group of heterogeneous syndromes in which an underlying neurodegenerative cause results in language deficits as the most salient feature (Mesulam, 2001). Current consensus criteria define three variants of PPA: semantic variant (svPPA), logopenic variant (lvPPA), and nonfluent or agrammatic variant (nfvPPA) (Gorno-Tempini et al., 2011). SvPPA is associated with asymmetrical, left greater than right anterior temporal lobe atrophy (e.g. Rohrer et al., 2010). Impaired naming and single-word comprehension deficits are identified as core criteria for svPPA (Gorno-Tempini et al., 2011). LvPPA features atrophy in the left posterior perisylvian and temporoparietal regions, including the superior and middle temporal gyri (e.g. Leyton, Piguet, Savage, Burrell, & Hodges, 2012). Word retrieval difficulty and impaired sentence repetition are identified as core criteria for lvPPA (Gorno-Tempini et al., 2011). NfvPPA is associated with left inferior frontal lobe, insula, and superior temporal lobe atrophy (Rohrer et al., 2010). Compromised grammar and/or motor aspects of speech are identified as core criteria for nfvPPA (Gorno-Tempini et al., 2011). None of the phenotypic variants excludes AD as the possible underlying neurodegenerative cause, however, lvPPA has been reported to have substantially higher probability of AD in comparison to svPPA and nfvPPA (e.g. Leyton et al., 2011).

Neuropathological diagnostic criteria for AD are based on tau-related pathology, which include neuritic plaques and neurofibrillary tangles (Hyman & Trojanowski, 1997), typically seen in the medial temporal lobe region, including the hippocampus, entorhinal cortex, and amygdala bilaterally (e.g. Desikan et al., 2009). The amnestic presentation is the most common syndromic presentation of AD, characterized by prominent deficits in learning and recall of information as

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https://doi.org/10.1016/j.bandl.2019.03.001
Received 17 August 2018; Received in revised form 20 February 2019; Accepted 18 March 2019
Available online 03 April 2019
0093-934X/

well as impairment in at least one other cognitive domain (McKhann et al., 2011). In the most recent diagnostic guidelines, McKhann et al. (2011) define three additional nonamnestic presentations of AD: language, visuospatial, and executive dysfunction. Core features of the nonamnestic presentations include word-finding difficulties, impaired spatial cognition, and deficits in reasoning, judgment and problem solving, respectively.

Since the diagnostic criteria for PPA were published (Gorno-Tempini et al., 2011), there have been several reports of unclassifiable cases (e.g. Sajjadi, Patterson, Arnold, Watson, & Nestor, 2012). In a study of 46 patients with PPA, Sajjadi et al. (2012) found that 23.3% fitted svPPA, 26.1% fitted nfvPPA, and 4.3% fitted lvPPA, while 41.3% were unclassifiable. The unclassified cases typically involved patients with diffuse deficits that did not apply to one variant and commonly fulfilled the criteria for multiple variants. Hoffman, Sajjadi, Patterson, and Nestor (2017) applied a data-driven approach to the PPA cohort reported by Sajjadi et al. (2012), which disregarded current diagnostic criteria. Clustering clearly separated svPPA patients, however, no clear classification was found for lvPPA or nfvPPA. Sajjadi, Patterson, and Nestor (2014) found that 14 of the unclassified PPA patients from the same cohort showed left temporoparietal atrophy, an atrophy pattern consistent with lvPPA. Unlike the classification criteria for lvPPA, these patients showed deficits in visuospatial processing, working memory, and episodic memory, in addition to phonologic, morphosyntactic, and semantic deficits in language function. In the majority of PPA neuropathological and biomarker studies, lvPPA has been associated with an atypical presentation of AD (e.g. Leyton et al., 2011). In a biomarker study of 19 patients diagnosed with lvPPA, Teichmann et al. (2013) found that two-thirds of patients revealed a profile indicative of AD pathology. PPA caused by AD is atypical due to the asymmetric left hemisphere-dominant distribution of plaques and tangles (Mesulam et al., 2014). For use in research, Vandenberghe (2016) proposed that lvPPA encompasses two subtypes; left hemisphere-dominant AD lvPPA, closely related to linguistic AD, and phonological working memory deficit with focal damage of temporoparietal transition zone lvPPA, reflecting the original diagnostic criteria recommended by Gorno-Tempini et al. (2004).

Although impaired sentence and phrase repetition is a core feature of lvPPA (Gorno-Tempini et al., 2011), its significance as a diagnostic criterion remains poorly understood. Moreover, Mesulam and Weintraub (2014) proposed that the presence of repetition impairments should be an optional feature in lvPPA classification. With an aim to investigate the underlying cognitive mechanisms of repetition deficits in PPA and AD, Leyton et al. (2014) assessed 63 patients (13 svPPA, 10 lvPPA, 17 nfvPPA, and 23 amnestic AD) on the sentence repetition task from the Multilingual Aphasia Examination (Benton & Hamsher, 1989). Leyton et al. (2014) found that all diagnostic groups showed impaired performance on the sentence repetition task in comparison to healthy controls, based on the proportion of correctly repeated sentences, with no significant differences found between diagnostic groups. These findings are inconsistent with Foxe, Irish, Hodges, and Piguet (2013), and Meyer, Snider, Campbell, and Friedman (2015), who found that patients with lvPPA were significantly more impaired than AD patients on sentence repetition. Comparisons with svPPA and nfvPPA were not made in these studies. Leyton et al. (2014) proposed that breakdown in different cognitive systems account for impaired sentence repetition across the diagnostic groups. Specifically, sentence repetition impairment in lvPPA is due to phonological storage deficits, executive deficits in AD, impaired rehearsal abilities in nfvPPA, and impaired semantic knowledge in svPPA. These hypotheses reflect theoretical accounts of repetition impairments in the literature. Specifically, disruption of phonological short-term memory has been linked to the left temporoparietal junction (Hickok & Poeppel, 2007), consistent with atrophic change seen in lvPPA. It has been suggested that repetition and working memory deficits in lvPPA may overlap with impairments seen in some cases of AD, given the potential connection in underlying pathology (Tree & Kay, 2015). Attention/executive deficits (Leyton et al., 2012) and lexical/semantic processing deficits (Caza & Belleville, 2008) have also been proposed to impact verbal short-term memory capacity in AD. Semantic memory has also been reported to play a critical role in verbal short-term memory capacity in svPPA (e.g. Jefferies, Patterson, Jones, Bateman, & Ralph, 2004). These theoretical accounts are predominantly based on immediate serial recall tasks of number and non-number words.

To investigate the use of sentence repetition as a core feature of lvPPA, Hohlbaum et al. (2018) developed an error classification schema to evaluate performance beyond correctness judgments. Eight participants diagnosed with lvPPA were assessed on a sentence repetition task created by Friedmann and Grodzinsky (1997), forward digit span (Wechsler Memory Scale-III; WMS-III; Wechsler, 1997), and the Corsi Block-Tapping Test (Corsi, 1972). Significant differences were found between the control group and the lvPPA group for phonological errors, semantic substitutions, and formal errors (i.e. words that resulted in a similar phonological word form but were a different word). Furthermore, a significant difference between the frequency of errors in long and short sentences was found, indicative of reduced performance with increased sentence length. Phonological errors and omissions were the most frequently occurring errors for lvPPA. No correlation analyses were performed with sentence repetition performance. Given that all diagnostic groups in the studies reported above showed impairment on the sentence repetition task, the comparison of PPA variants and AD with respect to sentence repetition errors is warranted.

Immediate serial recall tasks, typically digit spans, are commonly reported with sentence repetition outcomes. In the comparison of diagnostic groups, Leyton et al. (2014) assessed forward and backward digit span (Wechsler, 1997). On the digit span tasks, svPPA were the only group to show normal performance, with no significant differences found between the performance of lvPPA, nfvPPA, and AD groups. Foxe et al. (2013) reported impaired performance on digit span tasks for lvPPA and AD groups, however, lvPPA patients performed significantly worse than AD patients. Foxe et al. (2013) reported that both lvPPA and AD group sentence repetition performance correlated with digit span performance, specifically, forward and backward subtests for AD patients and forward only for lvPPA patients. These findings are consistent with Small, Kemper, and Lyons (2000) who observed strong correlations between performance on digit span tasks and sentence repetition for AD patients. Small et al. (2000) proposed that the resource capacity theory (i.e. the limited pool of activation resources that accommodates the storage and computational demands of verbal information processing) provides an explanation in that sentence repetition and digit span tasks both draw upon a common pool of resources. Such theoretical accounts of working memory capacity, largely focused around primary memory, secondary memory, and attention control (i.e. inhibition), endorse a system for attention to memory, rather than working memory as a memory system itself (Oberauer, Süβ, Wilhem, & Sander, 2007). Specifically, primary memory is often construed as a limited capacity storage, representative of a person's attentional focus, typically 3–5 items (e.g. Cowan, 2010). In contrast to theories of primary memory as a storage system, some accounts propose a binding system in which capacity is not fixed, but rather, is determined by an individual's ability to form and break temporary associations between relevant memory units (e.g. Oberauer et al., 2007). Accordingly, differences in individual working memory capacity can be explained through the ability to form and break temporary associations. For working memory tasks that require maintenance of more than 3–5 items, retrieval from secondary memory is required (i.e. relevant memory units are displaced from primary memory and therefore demand longer term storage). The interaction of maintenance in primary memory and retrieval from secondary memory underpins the dual-component model of working memory capacity (Unsworth, Spillers, & Brewer, 2009). This dynamic interaction of working memory capacity has been found to be strongly associated with attention control (Shipstead, Lindsey, Marshall, &

Engle, 2014). In contrast to fixed-and maintenance-related working memory capacity theories, attention control associates working memory capacity with a person's ability to focus on contextually relevant information and resist distractions from environmental or cognitive events, thereby allowing effective retention of memory units (Kane, Conway, Hambrick, & Engle, 2007). Given the lack of understanding regarding the nature of phonological working memory deficits in lvPPA, specifically deficits of storage, rehearsal, and/or buffer processes (Foxe et al., 2013), interpretation using the dual-component model of capacity is warranted, with needed comparisons to other dementia syndromes.

This study aimed to investigate sentence repetition in people with different dementia syndromes, using an error classification schema adapted from Hohlbaum et al. (2018), and explore correlations with digit span abilities, tasks known to draw on verbal working memory. To our knowledge, no study has directly compared error patterns on sentence repetition tasks in svPPA, lvPPA, and AD participants. We hypothesized that lvPPA participants would show significantly reduced overall correctness in sentence repetition, in comparison to svPPA and AD. We further predicted that diagnostic groups would differ in quantity and type of errors. Specifically, we anticipated that phonological errors would only be seen for lvPPA participants and related word substitution errors would only be seen for svPPA participants, attributable to phonological loop and semantic memory deficits, respectively. Furthermore, we predicted error patterns for AD participants would be characterized by unrelated word substitution errors, attributable to attention control deficits. We hypothesized that lvPPA participants would show significantly reduced digit span performance in comparison to svPPA and AD participants. Finally, we hypothesized that digit span performance would be positively correlated with sentence repetition for all participants (across diagnostic groups).

2. Method

2.1. Participants

Twelve adults, aged between 55 and 86 years ($M = 65.5$), were recruited via private neurologists, geriatricians, and speech-language pathologists working in Perth, Australia (see Table 1 for demographic information and performance on neuropsychological tests for diagnostic groups). Participants were diagnosed based on the current international consensus criteria (Gorno-Tempini et al., 2011) and McKhann's diagnostic guidelines for AD (McKhann et al., 2011). All AD participants presented with impaired learning and information recall, as well as word-finding difficulties, however, clinical presentation was not classified further. Participants were native English-speaking and had no reported hearing impairments. Brain imaging, specifically MRI and FDG-PET, are provided in Figs. 1 and 2. Individual data for performance on neuropsychological tests are provided in the supplementary material.

2.1.1. General language and cognitive assessment

All participants were tested on a language and cognitive battery. The assessment battery comprised the Addenbrooke's Cognitive Examination third edition, ACE-III (Mioshi, Dawson, Mitchell, Arnold, & Hodges, 2006), Stroop Tests (Stroop, 1935), Trial Making Test, TMT (Tombaugh, 2004), Pyramids and Palm Trees Test, PPTT (Howard & Patterson, 1992), and the confrontation naming and word and non-word repetition subtests of the Northwestern Naming Battery, NNB (Thompson & Weintraub, 2014).

Insert Table 1

2.1.2. svPPA

Participant 1 (P1) was a 62 year-old man who reported a 5-year history of word-finding difficulties. He was working as a metallurgist at the time of the study. Participant 2 (P2) was a 55 year-old man who reported a 4-year history of word-finding difficulties and single word comprehension deficits. Due to the communication demands of his job as an engineer, he retired 2 years prior to the initiation of this study. Participant 3 (P3) was a 72 year-old man who reported a 12-year history of word-finding difficulties, comprehension difficulties, and more recent writing difficulties. He previously worked as a director of children's homes. Participant 4 (P4) was a 72 year-old man who reported a 2-year history of word-finding difficulties. He was working as a farmer at the time of the study, having retired from his previous position in the computer business.

On the Stroop tests, P2 and P3 showed marked difficulty, likely impacted by their impaired semantic memory. P2 and P3 showed moderate deficits on the TMT, while P1 and P4 showed normal performance on both. P1 showed mild deficits on the PPTT, while P2 and P3 showed marked impairment. Noun conceptual semantics was within normal limits for P4. P2 and P3 showed marked impairment on noun and verb confrontation naming, while mild deficits for nouns only were seen for P1 and P4. Ceiling performance for non-word and word repetition was seen for all svPPA participants.

Examination of P1's MRI showed prominent asymmetric atrophy in the left temporal lobe with widening of the temporal sulci and the left temporal horn. P1's FDG-PET showed markedly reduced activity in the left temporal lobe, most marked in the anterior pole. P2's MRI revealed striking atrophy of the left temporal lobe, most prominent anteriorly, congruent with the FDG-PET, which showed prominent hypometabolism in the left temporal lobe. P3's MRI revealed generalized supratentorial atrophic change with more prominent involvement of the anterior and mesial temporal lobe structures on the left. P3's FDG-PET showed markedly reduced activity in the left anterior temporal lobe. P4's MRI revealed atrophy of the left temporal lobe, consistent with marked hypometabolism in the left temporal lobe found on the FDG-PET.

2.1.3. lvPPA

Participant 5 (P5) was a 60 year-old female who reported a 3-year history of difficulties pronouncing words and word-finding difficulties. Due to the communication demands of her job as a deputy principal, she retired 2 years prior to this study. Participant 6 (P6) was a 70 year-old ex-army man who presented with a 4-year history of pronunciation and word-finding difficulties. Participant 7 (P7) was a 64 year-old female who reported a 2-year history of word-finding difficulties. She previously worked as a cleaner. Participant 8 (P8) was a 59 year-old female who reported a 2-year history of difficulties pronouncing words and word-finding difficulties. Due to increasing language difficulties, she retired from her work in customer service.

All lvPPA participants showed difficulty completing at least one of the Stroop Tests. On the TMT, marginal deficits were found for P5, while moderate deficits were found for the other 3 participants. Normal performance on the PPTT was found for all participants. Mild deficits in verb confrontation naming were seen for P6 and P8. P5 and P6 showed mild deficits on non-word repetition, while ceiling performance of word repetition was seen for all participants.

Examination of P5's MRI revealed some asymmetric loss of volume within the left superior temporal gyrus with mild prominence of the adjacent part of the cistern and superior temporal sulcus. P5's FDG-PET showed moderate to severe hypometabolism in the left temporal lobe, most marked at the superior and lateral aspects. P6 showed posterior temporoparietal atrophy with generalized mild volume loss in the perisylvian region. P6's FDG-PET showed markedly reduced activity in the parietal and temporal lobes, more severe on the left side compared to the right, and superior left frontal lobe. P7's MRI showed left anterior temporal lobe atrophy, congruent with the FDG-PET, which showed hypometabolism in the left anterior temporal lobe. P8's MRI revealed mild prominence of the extra-axial sulcal spaces around the convexities bilaterally and around the temporal lobes. P8's PFG-PET showed marked hypometabolism in the temporal, parietal, and occipital lobes,

Table 1
Demographic information and performance on neuropsychological tests for diagnostic groups.

	Diagnosis						
	svPPA		lvPPA		AD		
	Mean	Std dev	Mean	Std dev	Mean	Std dev	Normative data *M (SD)*
Age (years)	65.3	7.2	63.3	4.3	68.0	11.0	
Education (years)	14.5	3.0	12.3	2.9	12.5	2.5	
Time post diagnosis (years)	2.3	0.8	2.0	1.0	2.8	1.3	
ACE – III (/100)	53.8	25.8	61.8	12.9	39.5	15.9	58.7(21.4)[1]
Attention (/18)	14.3	3.9	13.8	3.5	9.5	4.3	13.8(4.4)[1]
Memory (/26)	14.3	4.9	9.8	3.5	6.8	3.5	–
Fluency (/14)	5.0	2.8	7.5	3.0	2.3	0.8	–
Language (/26)	11.5	9.7	18.0	1.9	14.0	4.2	12.8(6.3)[1]
Visuospatial (/16)	13.5	2.1	12.8	3.4	7.0	4.9	13.3(2.8)[1]
Stroop							
Word score	63.5	29.7	80.0	12.9	39.5	19.2	103.8[2]
Colour score	41.5	18.6	45.3	7.3	19.5	10.2	80.6[2]
Colour-word score	38.0	2.0	14.0	8.5	11.0	4.3	48.4[2]
TMT – part A (seconds)	52.3	18.3	49.3	15.9	163.3	47.2	35.7(12.8)[3]
TMT – part B (seconds)	122.5	57.9	241.0	106.1	350.0	0.0	81.5(36.1)[3]
PPTT (/52)	38.5	7.8	50.8	1.3	36.8	8.0	47.6(2.6)[4]
NNB							
Confrontation naming nouns (/16)	5.5	5.5	5.5	5.5	5.5	5.5	12.1(5.3)[5]
Confrontation naming verbs (/16)	9.5	5.1	9.5	5.1	9.5	5.1	12.6(3.9)[5]
Non-word repetition (/10)	10.0	0.0	9.0	1.0	8.8	2.2	8.6(1.9)[5]
Word repetition (/21)	21.0	0.0	21.0	0.0	21.0	0.0	20.6(1.0)[5]

ACE-III: Addenbrooke's Cognitive Examination third edition, PPTT: Pyramids and Palm Trees Test, NNB: Northwestern Naming Battery.

[1] Normative data for PPA (Hsieh, Schubert, Hoon, Mioshi, & Hodges, 2013. Cut-off at overall score 82 indicates 100:1 likelihood of dementia Mioshi et al., 2006).

[2] Normative data mathematically estimated from total time per hundred words to number of times in 45 s (Van der Elst, Van Boxtel, Van Breukelen, & Jolles, 2006).

[3] Normative data based on healthy ageing sample (age $M = 72.4$, $SD = 8.5$). AD normative data; part A – 67.1(31.0), part B – 190.8(81.6) (Ashendorf et al., 2008).

[4] Normative data based on healthy sample aged 20–63 (Mansur, Carthery-Goulart, Bahia, Bak, & Nitrini, 2013).

[5] Adapted from the NNB normative data for 37 individuals with PPA aged 48–81 (Thompson & Weintraub, 2014).

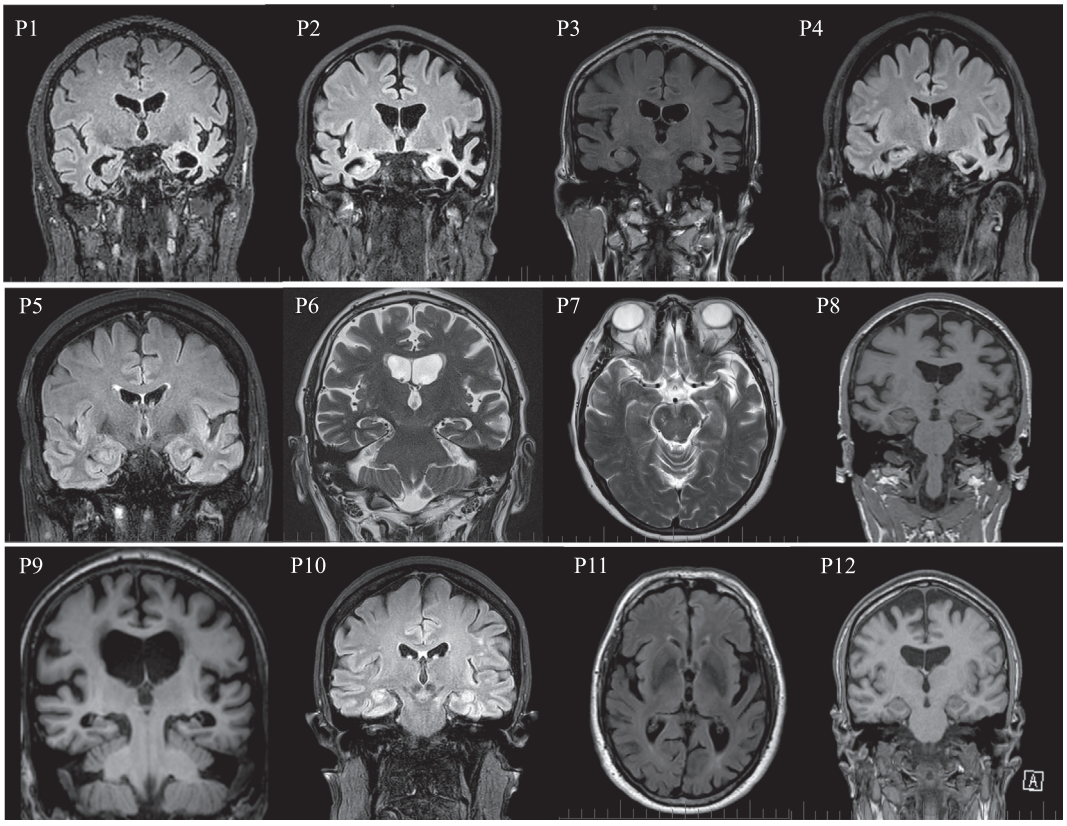

Fig. 1. MRI scans of participants diagnosed with svPPA (P1, P2, P3 and P4), lvPPA (P5, P6, P7 and P8), and AD (P9, P10, P11 and P12).

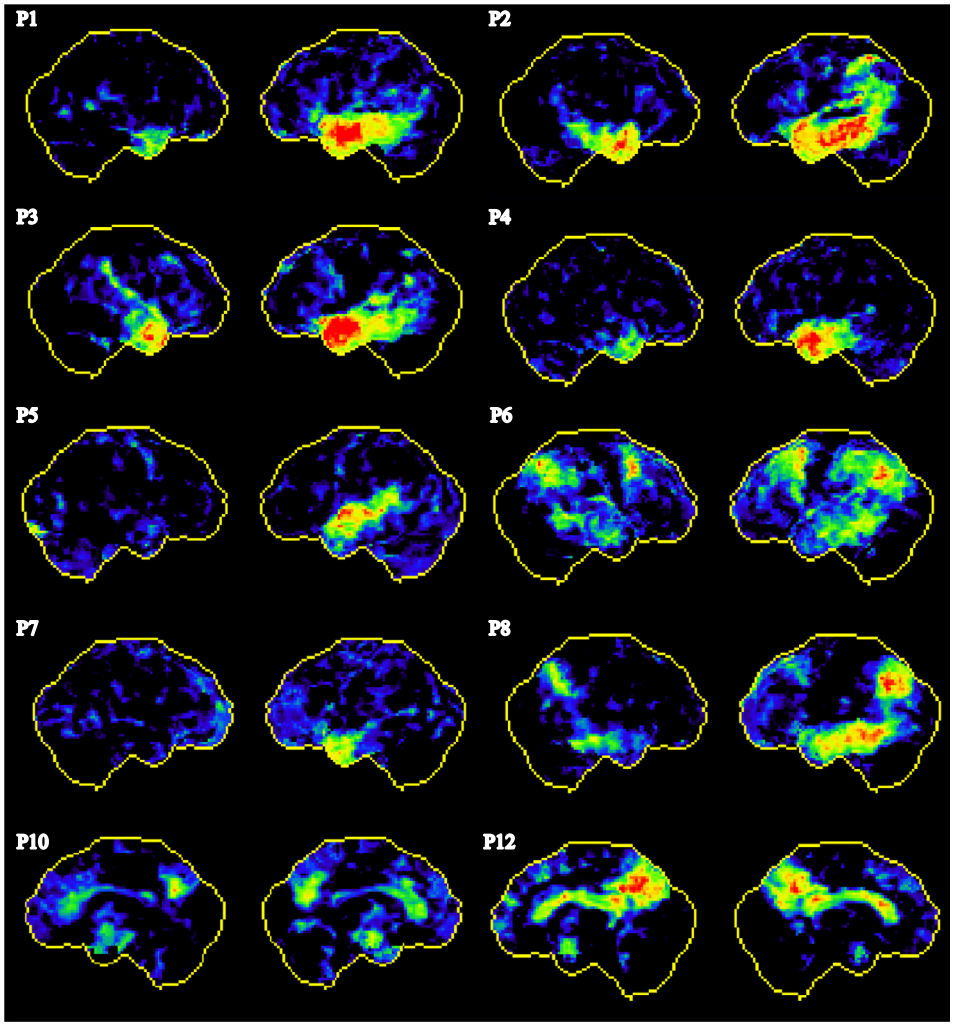

Fig. 2. FDG-PET scans of participants diagnosed with svPPA (P1, P2, P3 and P4), lvPPA (P5, P6, P7 and P8), and AD (P10 and P12).

with left hemisphere dominance.

2.1.4. AD

Participant 9 (P9), a retired bank teller, was an 86 year-old man who reported a 4-year history of impaired short-term memory, orientation, and word-finding difficulties despite intact comprehension. Participant 10 (P10) was a 60 year-old female who reported a 2-year history of short-term memory and word-finding difficulties, as well as orientation and comprehension deficits. Due to increasing memory and language difficulties, she retired from her work as a garden and nursery worker. Participant 11 (P11), a retired real-estate agent, was a 68 year-old man who reported a 5-year history of visuo-spatial processing and short-term memory difficulties, as well as impaired comprehension and occasional word-finding difficulties. Participant 12 (P12) was a 58 year-old male who reported a 6-year history of impaired comprehension, short-term memory, visuo-spatial processing, and occasional word-finding difficulties. Due to the communication demands of his job as a geologist, he retired 5 years prior to this study.

Moderate deficits were seen on the Stroop Tests and the TMT for participants with AD, with marked difficulty seen for P12. Marked impairments on the PPTT were found for P11 and P12, while P9 and P10 showed mild deficits. Deficits on confrontation naming, particularly verbs, were found for all participants with the exception of P10 who performed at ceiling level. Intact non-word and word repetition was found for all participants with the exception of P11 who showed moderate impairment on the non-words subtest.

Examination of P9's MRI revealed mild to moderate symmetric cerebral hemisphere atrophic change. P10's MRI showed minimal generalized involutional change with early sulcal widening over both convexities, considered normal for the participant's age. P10's FDG-PET revealed reduced cortical metabolism of the frontal lobes and parietal lobes, with involvement of the precuneus and the anterior cingulate, bilaterally. P11's MRI showed generalized involutional changes with sulcal widening. P12's MRI showed generalized cerebral hemisphere involutional change, most prominent with the frontal and parietal regions, congruent with the FDG-PET, which showed reduced activity at the site of atrophy on the MRI with involvement of the precingulate gyrus and the precuneus. No FDG-PET results were available for P9 or P11.

2.2. Procedure

Participants were examined in one to three sessions at intervals of two to three days. Examinations were completed within the participant's home with the exception of P4 who lived in rural Western Australia and was assessed in clinic rooms in Perth. Ethics approval was received from the Curtin University Human Research Ethics Committee (HR218/2015).

2.2.1. Assessment of sentence repetition

The repetition task from the Arizona Battery for Communication Disorder of Dementia (ABCD) (Bayles & Tomoeda, 1993) was used to

examine sentence repetition performance. The assessment contains 10 phrases/sentences made up of real words that are grammatically correct, however, are not semantically or syntactically meaningful. In the items in this test, all words used are nouns, verbs, and adjectives. The task items comprised six and nine syllable sentences. Prior to administering the repetition task, the instructions were provided by the examiner (first author) in accordance with the test manual. The examiner presented each item with natural pronunciation and intonation. Repetition of the target sentence was not permitted, however, the examiner recorded when the participant requested the item be repeated. All 10 items were administered for each participant, despite manual instructions to discontinue after the first five items if an error occurred.

2.2.2. Sentence repetition error evaluation schema

In order to evaluate the performance of each participant on the sentence repetition test, a classification schema was developed to evaluate performance, adapted from Hohlbaum et al. (2018).

At the *sentence level*, each repetition item was evaluated for the following:

- *Overall correctness,* each sentence was scored based on the number of syllables immediately correct (i.e. without error or delayed self-correction)
- *Stimulus repeat request*, each request made by a participant for repetition of the stimulus was recorded, however, no repetitions were provided
- *Incorrect word order,* sentences which contained incorrect word order.

At the *word level*, each repetition item was evaluated for the following:

- *Related word substitution,* words substituted with a semantically related word
- *Unrelated word substitution,* words substituted with a semantically unrelated word
- *Beginning omission,* omission of word/s at the beginning of a sentence
- *Middle omission,* omission of word/s in the middle of a sentence
- *Ending omission,* omission of word/s at the end of a sentence
- *Phonological error,* erroneous words due to phonological processes (e.g. phoneme substitution, omission, addition, or deletion)
- *Morpho-phonological error,* erroneous words due to grammatical morphemes altered by phonological errors.

2.2.3. Assessment of digit span

Digit span was assessed using the digit span subtest of the WMS-III (Wechsler, 1997). Specifically, total digit span (i.e. combined score of forward and backward digit span), forward digit span, and backward digit span scores were determined.

2.3. Statistical analyses

The research aims were addressed with a series of Generalized Linear Mixed Models (GLMMs). All GLMMs were implemented through SPSS's 24.0 GENLINMIXED procedure. The GLMM represents a special class of regression model. The GLMM is 'generalized' in the sense that it can accommodate dependent variables with markedly non-normal distributions; the GLMM is 'mixed' in the sense that it includes both random and fixed effects. Two types of GLMMs were tested. Time post diagnosis was not a confounding in any of the GLMMs and was therefore not included as a covariate in any of the models.

A series of GLMMs was tested in order to determine the effect of diagnosis on overall correctness on sentence repetition and error patterns (stimulus repeat request, incorrect word order, related word substitution, unrelated word substitution, beginning omission, middle omission, ending omission, phonological error, and morpho-phonological error). Each of the GLMMs included one nominal random effect (participant) and one nominal fixed effect (diagnosis: svPPA, lvPPA, AD). If a diagnostic group shows zero variance in a particular sentence repetition error pattern, the cell will be omitted from further analysis. This model was also used to analyze the effects of diagnosis on total digit span, forward digit span, and backward digit span. The traditional ANOVA between-groups model requires the following assumptions to be satisfied: normality and homogeneity of variance. The GLMM 'robust statistics' option was invoked to accommodate any violations of these assumptions.

A single GLMM was tested in order to determine whether total digit span, forward digit span, and backward digit span predicted the sentence repetition overall correctness. The GLMM included one nominal random effect (participant) and three ordinal fixed effects (total digit span, forward digit span, and backward digit span). The traditional linear regression model requires the following assumptions to be satisfied: normality of residuals, linearity, and homoscedasticity. The GLMM 'robust statistics' option was invoked to accommodate any violations of these assumptions.

Table 2
Raw scores (scaled scores in brackets) for sentence repetition and digit span tasks for PPA and AD participants.

	P1	P2	P3	P4	P5	P6	P7	P8	P9	P10	P11	P12
Diagnosis	svPPA	svPPA	svPPA	svPPA	lvPPA	lvPPA	lvPPA	lvPPA	AD	AD	AD	AD
ABCD[1]												
Sentence repetition (/75)	56	42	58	67	24	44	60	26	53	19	21	36
Sentence repetition error analysis												
Stimulus repeat request	0	1	0	0	0	0	0	2	0	2	2	0
Incorrect word order	0	0	0	0	0	0	0	0	0	0	0	0
Related word substitution	1	0	0	1	2	0	0	0	1	1	0	0
Unrelated word substitution	0	0	0	0	1	0	2	2	0	7	3	0
Beginning omission	0	0	0	0	0	0	0	0	0	0	0	0
Middle omission	3	6	5	2	0	2	0	0	0	0	0	0
Ending omission	2	3	0	1	8	5	2	4	4	6	7	7
Phonological error	0	0	0	0	4	3	2	0	0	0	0	0
Morpho-phonological error	0	0	0	0	2	2	0	0	0	0	1	1
WMS – III[2]												
Digit span forward (/16)	9	7	9	13	5	9	11	7	11	8	4	5
Digit span backward (/14)	9	5	6	11	5	4	2	5	7	3	2	3
Digit span overall (/30)	18 (11)	12 (7)	15 (10)	24 (17)	10 (6)	13 (8)	13 (8)	12 (7)	18 (14)	11 (7)	6 (4)	8 (5)

[1] ABCD: Arizona Battery for Communication Disorders of Dementia. Performance Means and SDs for old normal controls (*M* age: 74 years, *SD*: 5.4) scored *M*: 67.9 (*SD*: 7.0).

[2] WMS-III: Wechsler Memory Scale third edition.

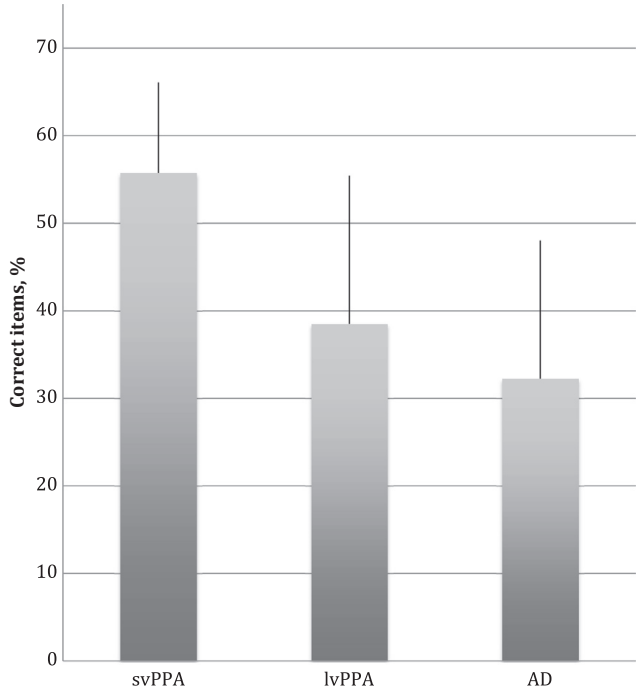

Fig. 3. Performance on sentence repetition across groups. SvPPA, semantic variant; lvPPA, logopenic variant; AD, Alzheimer's disease. Bars show standard error.

3. Results

Raw scores for individual participants for sentence repetition performance, error analysis on sentence repetition, and digit span performance are presented in Table 2.

3.1. Performance on sentence repetition

Overall correctness on the sentence repetition task was scored for each participant based on the number of syllables immediately correct. All participants demonstrated compromised performance on this task (see Fig. 3). Across the three diagnostic groups, a significant group effect was found ($f = 6.005$; $df = 2,8$; $p = 0.026$). Further analyses through pairwise comparisons revealed significant differences in overall correctness between svPPA and lvPPA ($p = 0.036$), as well as svPPA and AD ($p = 0.022$), indicating that participants with svPPA performed significantly better on the sentence repetition task than lvPPA and AD participants. No significant difference in overall sentence repetition performance was found between lvPPA and AD participants.

3.2. Error patterns on sentence repetition

For the error pattern unrelated word substitution, zero variance was found for svPPA. A pairwise comparison between lvPPA and AD revealed no significant differences in unrelated word substitution errors. For the error pattern middle omission, zero variance was found for AD. A pairwise comparison between lvPPA and svPPA revealed a significant difference between group ($f = 15.077$; $df = 1,6$; $p = 0.008$), indicating that middle omissions occurred significantly more for svPPA than lvPPA participants. For ending omissions, a significant group effect was found ($f = 15.268$; $df = 2,9$; $p = 0.001$). Further analyses through pairwise comparisons revealed significant differences between lvPPA and svPPA ($p = 0.026$), as well as AD and svPPA ($p = < 0.001$), indicating that ending omissions occurred significantly more for participants with lvPPA and AD in comparison to svPPA participants. No significant group effects were found for stimulus repeat request, related word substitution, unrelated word substitution, and morpho-phonological errors, with all four error patterns occurring either infrequently or not at all for each diagnostic group. Phonological errors were only found for participants diagnosed with lvPPA, therefore, this error pattern was not amenable to statistical analysis. Incorrect word order and beginning omission had zero variance in all three diagnostic groups, therefore, were not amenable to statistical analysis (see Fig. 4).

3.3. Performance on digit span

Total digit span was scored for each participant as per the WMS-III instructions (Wechsler, 1997). All participants performed within normal limits on total digit span, with the exception of P11 and P12 (both AD). Across the three diagnostic groups, a significant group effect was found ($f = 8.408$; $df = 2,8$; $p = 0.011$). Further analysis through pairwise comparisons showed significant differences between svPPA and lvPPA ($p = 0.009$), as well as svPPA and AD ($p = 0.008$), indicating

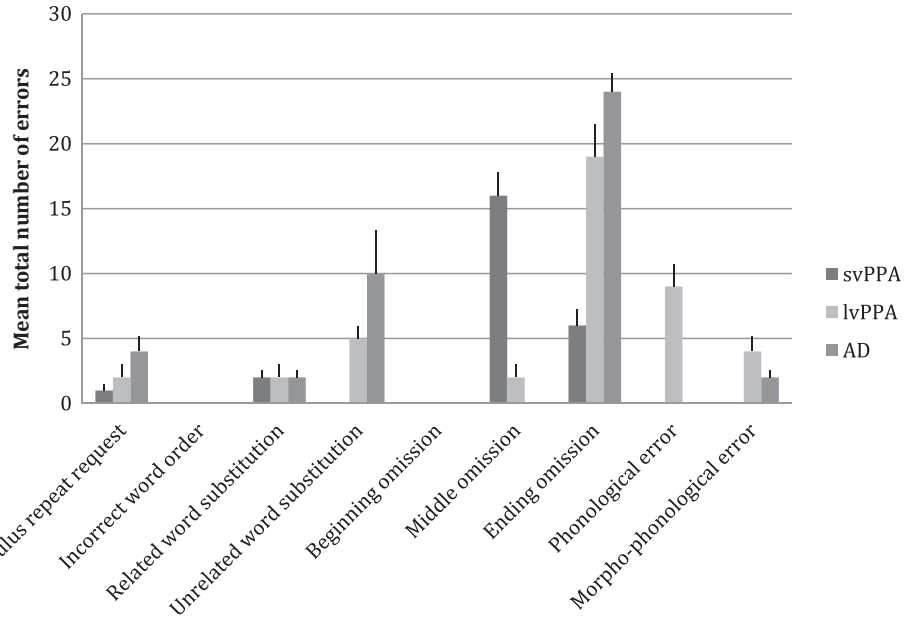

Fig. 4. Error patterns on sentence repetition across groups. SvPPA, semantic variant; lvPPA, logopenic variant; AD, Alzheimer's disease. Bars show standard error.

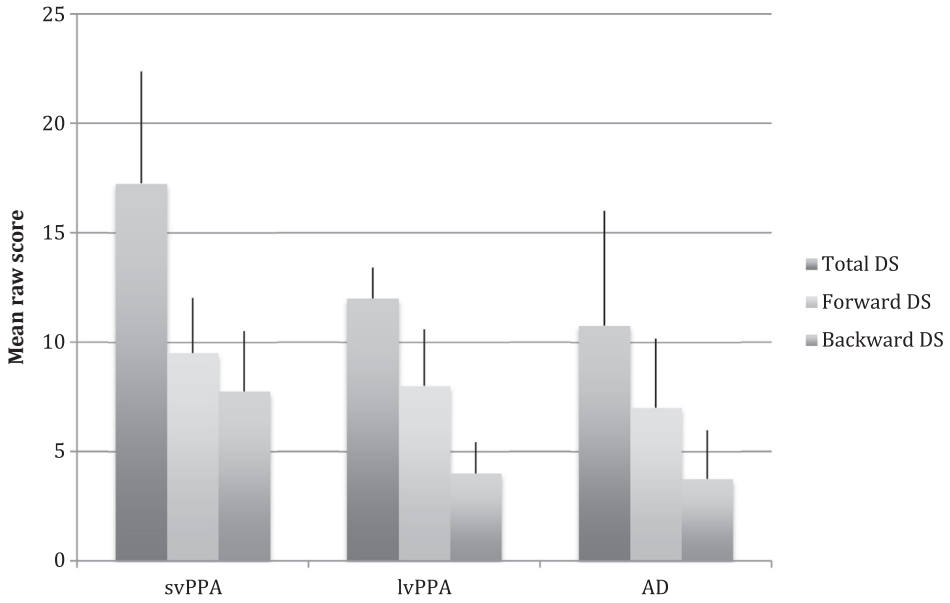

Fig. 5. Performance on digit span across groups. SvPPA, semantic variant; lvPPA, logopenic variant; AD, Alzheimer's disease. Bars show standard error.

that participants with svPPA performed significantly better on the digit span subtest, compared to lvPPA and AD participants. No significant difference in overall digit span performance was found between lvPPA and AD participants (see Fig. 5).

All participants performed within normal limits on forward digit span, with the exception of P11 who showed borderline impairment (e.g. Howieson, Holm, Kaye, Oken, & Howieson, 1993). Across the three diagnostic groups, no significant group effect ($f = 3.500$; $df = 2,8$; $p = 0.081$) was found for performance on forward digit span, indicating no significant differences between diagnostic groups. The average score for lvPPA ($M = 7.395$) was lower than svPPA ($M = 9.349$) and AD ($M = 7.756$), however, these differences were not found to be significant.

For backward digit span, all participants diagnosed with svPPA and lvPPA performed within normal limits, with the exception of P7 who showed impaired performance. Three of the four participants diagnosed with AD showed impaired performance on backward digit span, with the exception of P9 who performed within normal limits. Across the three diagnostic groups, a significant group effect was found ($f = 7.695$; $df = 2,8$; $p = 0.014$) for performance on the backward digit span task. Further analyses through pairwise comparisons showed significant differences between svPPA and lvPPA ($p = 0.011$), as well as svPPA and AD ($p = 0.009$), indicating that participants with svPPA performed significantly better on the backward digit span task than lvPPA and AD participants. No significant difference in the backward digit span task was found between lvPPA and AD participants.

3.4. Association between digit span and sentence repetition

A series of regression analyses were conducted to examine the correlation between digit span scores (i.e. total, forward, and backward digit span scores) and overall correctness on the sentence repetition task. Correlations were determined across diagnostic groups. The distribution was not normal, therefore Spearman's rho nonparametric test was used. A significant correlation was found between performance on the total digit span score and the sentence repetition task ($r_s = 0.847$, $p = 0.001$), indicative of a strong, positive correlation. A significant correlation was found between performance on the forward digit span and sentence repetition task ($r_s = 0.832$, $p = 0.001$), indicative of a strong, positive correlation. A non-significant correlation was found between performance on the backward digit span and sentence repetition task ($r_s = 0.512$, $p = 0.089$).

4. Discussion

This is the first study to investigat sentence repetition in people with different dementia syndromes, through a direct comparison of error patterns on sentence repetition tasks in svPPA, lvPPA, and AD participants. Correlations with digit span performance were explored to gain a greater understanding of how sentence repetition is mediated by working memory. In accordance with our hypotheses, performance on sentence repetition and digit span tasks varied across diagnostic groups, however, was not always consistent with our predictions. As expected, participants with svPPA had the greatest performance on overall correctness on sentence repetition in comparison to lvPPA and AD participants, although impairment was present and frequently characterized by middle omissions. In contrast, the most typically occurring error patterns in sentence repetition for lvPPA participants were ending omissions and phonological errors. For participants with AD, error patterns in sentence repetition also featured ending omissions as well as unrelated word substitutions. With regard to digit span, individuals diagnosed with svPPA had the highest performance on total and backward digit span in comparison to lvPPA and AD. By contrast, no significant differences between diagnostic groups were found for forward digit span. Lastly, significant correlations were found both for total digit span and sentence repetition, and forward digit span and sentence repetition, while an insignificant correlation was found for backward digit span and sentence repetition.

Impairments in sentence repetition were apparent for all diagnostic groups, although performance was significantly better for cases with svPPA. This finding does not support those of Leyton et al. (2014) who found no significant differences between groups. This disparity may be explained by potentially intact primary memory and impaired secondary memory in cases with svPPA. Specifically, in our study, the items on the sentence repetition task comprised up to 9 syllables whereas the Multilingual Aphasia Examination used by Leyton and colleagues consists of items up to 24 syllables. Consequently, svPPA participants may successfully rely on primary memory for items consisting of fewer syllables, but experience breakdown once retrieval from secondary memory is required. Alternatively, this could indicate an encoding breakdown rather than retrieval, warranting research utilizing both meaningful and non-meaningful words and sentences. Intact primary memory in cases with svPPA is further supported by performance within normal limits on all digit span tasks found in this study. Impaired retrieval from secondary memory in svPPA may be explained

by impaired use of ineffective rehearsal/recall strategies to retrieve information displaced in secondary memory. In order to effectively retrieve relevant memory units from the secondary memory, a person must search secondary memory on the basis of cues (e.g. associated information) (Unsworth & Engle, 2007). SvPPA patients may fail to retrieve memory units due to diffuse recall strategies as a result of impaired semantics (i.e. reduced ability to use cues such as associated information). Given that middle omissions, rather than ending omissions, were the most frequently occurring error pattern, there is an evident need to consider capacity theories other than storage, such as a binding system. Lack of related word substitution errors, inconsistent with our hypotheses, may suggest that svPPA participants did not use semantic associations to form binds in primary memory or use semantic-based retrieval cues in secondary memory. It is important, however, to consider the impact of non-meaningful sentences used by the Arizona Battery for Communication Disorder of Dementia and used in our study, which is likely to have reduced the opportunity for semantic associations and thus limit the potential occurrence of related word substitution errors. In addition, normal digit spans but impaired sentence repetition may support the role of semantic representations in verbal working memory. Jefferies et al. (2004) proposed that intact digit span, in comparison to impaired word span in svPPA patients, may be reliant on relatively preserved number knowledge. Moreover, Jefferies and colleagues attributed this to typically spared areas of the parietal lobes involved in numerical processing as well as the high frequency use of single-digit numbers in everyday life.

In contrast to our hypotheses, no significant differences in overall correctness on sentence repetition were found for lvPPA and AD groups. This finding is inconsistent with Foxe et al. (2013) who found that lvPPA patients were significantly more impaired than AD patients on sentence repetition, despite performance being compromised for both groups. Consistent with our hypotheses, phonological errors were only seen for lvPPA participants, although ending omissions were the most frequently occurring error pattern. These findings support Hohlbaum et al. (2018) who found that phonological errors and omissions were the most common errors for eight patients with lvPPA. The presence of phonological errors in the present study and Hohlbaum et al. (2018) study support previous research that propose verbal short-term memory deficits are a result of phonological disintegration (e.g. Leyton & Hodges, 2013). Thus, sentence repetition deficits appear to be critical to the diagnosis of lvPPA, however, the evaluation of phonological errors is likely critical for discrimination with AD. Similarly to lvPPA, ending omissions were most common for AD participants, followed by unrelated word substitutions. Although Small et al. (2000) did not systematically analyze errors in their study of people with AD, they reported that errors progressively increased from initial to medial to final portions of the sentence. Small and colleagues proposed that these errors denote a limited pool of resources that mediates verbal working memory and thus determines an individual's capacity for sentence repetition. Our findings of ending omissions as the most typical error for AD participants corroborate those of Small and colleagues. Given this high frequency of ending omissions and the fact that all lvPPA and AD participants performed within normal limits on forward digit span (with the exception of P11 who showed borderline performance), it is likely that participants may rely on primary memory for tasks within the fixed capacity of working memory, but experience breakdown once required storage exceeds this limit. For AD participants, primary memory capacity and successful retrieval from secondary memory may be impacted by attention control, i.e. the ability to resist distraction. People with poor attention control have been reported to lose focus due to distractions in the testing environment or cognitive events (e.g. mind-wandering) (McVay & Kane, 2011). Unrelated word substitutions may suggest that AD patients select irrelevant retrieval candidates due to ineffective searches of secondary memory, which may be due to poor attention control. Reduced attention control may also explain better performance on forward digit span than backward digit span for cases with AD, as backward span demands greater input of attention control due to the required manipulation of information. Unlike primary memory, secondary memory is subject to build up of interference in the retrieval phase therefore attention control is required (Craik & Birtwistle, 1971). Consequently, impaired attention control in AD may be further indicated by significantly impaired performance on the Stroop and TMT tasks, although the potential correlations of these attention and inhibitions tasks with sentence repetition and digit span were not analyzed in this study. Similar mechanisms of working memory capacity may account for ending omissions and phonological errors seen for lvPPA participants, however, alternative mechanisms could account for phonological errors. General consensus within the current lvPPA diagnostic literature indicates that impaired repetition is associated with deficits in the phonological loop (Gorno-Tempini et al., 2008), however, it remains unclear as to whether this is due to impairment of the storage, rehearsal and/or buffer processes of phonological working memory (Foxe et al., 2013). Through interpretation using the dual-component model, the presence of phonological errors may be explained through impaired retrieval from secondary memory due to impaired rehearsal processes, rather than ineffective or irrelevant search cues.

By looking beyond overall correctness scores on sentence repetition and applying an error evaluation schema, we have found evidence to support the heterogeneous nature of dementia syndromes, though this was most apparent for cases with svPPA in contrast to lvPPA and AD. Given the number of similarities found between lvPPA and AD diagnostic groups including overall correctness on sentence repetition, all error patterns (with the exception of phonological errors), and performance on all digit span tasks, this study reinforces the complexity of distinguishing between lvPPA and AD based on behavioural and cognitive profiling. The presence of phonological errors for cases with lvPPA in contrast to no phonological errors for cases with AD, mark the only outcome found to distinguish lvPPA and AD participants in this study. These findings may support the body of research that suggests that lvPPA may be a discrete AD endophenotype (e.g. Sajjadi et al., 2014), however, further evidence investigating the possibility of varying underlying mechanisms of working memory capacity in such dementia syndromes in warranted.

Significant correlations were found between both total digit span and sentence repetition, and forward digit span and sentence repetition, were not however found between backward digit span and sentence repetition. The significant correlations suggest that sentence repetition and forward digit span may place similar demands on the working memory system, reflective of the same mechanisms underlying an individual's working memory capacity in these tasks. In contrast, it is likely that backward digit span places different demands on working memory, ultimately providing a varying perspective of the cognitive processes that may define working memory capacity. Findings from this study indicate that cases with svPPA perform significantly better on backward digit span, providing further evidence to support different mechanisms underlying capacity which is relatively intact in svPPA in comparison to lvPPA and AD. Given the rehearsal component of backward digit span, it is hypothesized that this task places greater demand on attention control required to maintain information in primary memory, compared to forward digit span and sentence repetition. While sentence processing may be mediated by working memory (Small et al., 2000), our findings support the notion that multiple mechanisms are needed to explain individual as well as diagnostic differences in working memory capacity.

While this study has enabled preliminary application of a sentence repetition error evaluation schema across a small number of participants in considerable depth, the small sample size of the lvPPA, svPPA, and AD groups did impact the statistical power of the study. GLMMs were employed to minimise these effects as far as possible given their robust handling of small data sets. Furthermore, diagnoses in this study were not accompanied by postmortem confirmation or variant

diagnosis for cases with AD. The findings pose a number of questions for future research, with both theoretical and clinical impetuses. The findings from this study call for systematic investigation of both the cognitive and linguistic mechanisms that underlie verbal working memory capacity in different dementia syndromes. Specifically, application of a range of working memory tasks to these dementia syndromes is warranted, including but not limited to operation span, symmetry span, and running letter span tasks (see Shipstead et al., 2014 for further task information). In addition, research that includes a range of sentence repetition tasks, including contrasting performance on semantically meaningful sentences with the stimuli of the ABCD may provide useful information and, potentially, further confirm that including semantic relationships does not aid those with svPPA but may help lvPPA. Given that several different factors are considered here as the potential source of repetition impairments in each group, future research may also include stimuli sensitive to the linguistic and cognitive factors, such as length, complexity, and number of propositions within the sentence. Examination of multi-propositional sentences, as well as low- and high-predictability sentences may, too, further identify factors important to the relationship between sentence repetition tasks and different mechanisms in progressive language disorders. Finally, the inclusion of additional error patterns codes, for example, perseveration errors, may also further elucidate these relationships.

Our findings on sentence repetition and digit span tasks distinguish svPPA patients, however, no clear classification patterns were found for lvPPA and AD. Consequently, the diagnostic criteria of impaired sentence repetition may reflect varying perspectives of the cognitive processes that define this construct, which is further influenced by the task design. Based on the current findings, the use of sentence repetition tasks to aid differential diagnosis of dementia syndromes such as lvPPA and AD is likely limited, particularly without further application of error evaluation schemas to identify the presence or absence of phonological errors. Our findings show that the cognitive presentations and pathological processes of dementia syndromes such as PPA variants and AD may have significant implications for our theoretical understanding of the mechanisms underlying verbal working memory capacity and warrant in-depth investigation.

Statement of significance

Our findings on sentence repetition and digit span clearly separate svPPA participants, however, no clear classification patterns were found for lvPPA and AD. While sentence processing may be mediated by working memory, our findings support the notion that multiple mechanisms are needed to explain diagnostic differences in working memory capacity.

Declaration of interest statement

The authors report no declarations of interest.

Acknowledgments

This work was supported by the Dementia Australia Research Foundation and Curtin University through the Australian Government Research Training Program.

Appendix A. Supplementary material

Supplementary data to this article can be found online at https://doi.org/10.1016/j.bandl.2019.03.001.

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Chapter 4

Chapter 4 presents the findings of the second study in Phase Two of the research that aimed to examine the viability of connected speech as an intervention outcome measure, to inform the reliability for use in the evaluation of across level generalisation to functional contexts.

Study Overview

This study addressed a predominant assessment limitation identified in Phase One of the research program, specifically, the inadequate reporting of connected speech as an intervention outcome despite recognition of connected speech as a central component of language assessment in PPA and AD. As outlined in Chapter 1, researchers and clinicians encounter a range of challenges when planning to use connected speech and/or discourse outcome measures, specifically, the limited understanding of the reliability of task and genre selection, as well as consistency of successive sampling (Hird et al., 2006; Morello et al., 2017). Further barriers to the use of connected speech as an intervention outcome have been proposed, largely centered on the time intensive nature of sampling collection and evaluation. To inform the use of connected speech outcomes in both research and clinical practice, this study examined whether connected speech is was stable over successive sampling for the participant group of people with PPA and AD. The reliability of connected speech sampling, addressed through the degree of variability across samples collected within a specified period of time (e.g. multiple baseline sampling), requires investigation to inform use as an assessment tool to inform intervention and, subsequently, as a treatment outcome measure. Increased understanding of this outcome measure may increase accessibility and use as a measure of across level generalisation, critical for informing gains in language behaviours beyond the treatment level, and pertinent to assessment and intervention planning in Phase Three. To address this limitation, the stability of connected speech over successive sampling is examined on measures of lexical content, fluency, and communicative informativeness and efficiency.

Beales, A., Whitworth, A., Cartwright, J., Panegyres, P. K., & Kane, R. T. (2018). Determining stability in connected speech in primary progressive aphasia and Alzheimer's disease. *International Journal of Speech-Language Pathology*, *20*(3), 361-370.

International Journal of Speech-Language Pathology

ISSN: 1754-9507 (Print) 1754-9515 (Online) Journal homepage: https://www.tandfonline.com/loi/iasl20

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To cite this article: Ashleigh Beales, Anne Whitworth, Jade Cartwright, Peter K. Panegyres & Robert T. Kane (2018) Determining stability in connected speech in primary progressive aphasia and Alzheimer's disease, International Journal of Speech-Language Pathology, 20:3, 361-370, DOI: 10.1080/17549507.2018.1442498

To link to this article: https://doi.org/10.1080/17549507.2018.1442498

International Journal of Speech-Language Pathology, 2018; 20: 361–370

Determining stability in connected speech in primary progressive aphasia and Alzheimer's disease

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Abstract

Purpose: Using connected speech to assess progressive language disorders is confounded by uncertainty around whether connected speech is stable over successive sampling, and therefore representative of an individual's performance, and whether some contexts and/or language behaviours show greater stability than others.
Method: A repeated measure, within groups, research design was used to investigate stability of a range of behaviours in the connected speech of six individuals with primary progressive aphasia and three individuals with Alzheimer's disease. Stability was evaluated, at a group and individual level, across three samples, collected over 3 weeks, involving everyday monologue, narrative and picture description, and analysed for lexical content, fluency and communicative informativeness and efficiency.
Result: Excellent and significant stability was found on the majority of measures, at a group and individual level, across all genres, with isolated measures (e.g. nouns use, communicative efficiency) showing good, but greater variability, within one of the three genres.
Conclusion: Findings provide evidence of stability on measures of lexical content, fluency and communicative informativeness and efficiency. While preliminary evidence suggests that task selection is influential when considering stability of particular connected speech measures, replication over a larger sample is necessary to reproduce findings.

Keywords: *Primary progressive aphasia*; *Alzheimer's disease*; *connected speech*; *stability*; *elicitation genres and tasks*

Introduction

The evaluation of connected speech is becoming increasingly recognised as a useful diagnostic tool and context for measuring intervention outcomes. Primary progressive aphasia (PPA) and Alzheimer's disease (AD) are two such conditions (McKhann et al., 2011; Mesulam, 2001) where connected speech has assumed a recent focus as a context for better understanding the nature of the language features associated with progressive language disorders (Ash et al., 2013; Dijkstra, Bourgeois, Allen, & Burgio, 2004; Whitworth, Cartwright, Beales, Leitão, Panegyres, & Kane, 2017) and monitoring response to intervention (Whitworth et al., 2017). In a recent review, Boschi et al. (2017) included 61 papers reporting connected speech in the clinical diagnosis of neurodegenerative diseases. This included AD and three recognised variants of PPA: semantic variant (svPPA), logopenic variant (lvPPA) and nonfluent variant (nfvPPA). With regard to PPA, Boschi et al. (2017) reported 15 studies, 11 including individuals with svPPA, three including lvPPA, and one study involving nfvPPA. People diagnosed with svPPA were reported to present with simplification of syntax, impaired local coherence, as well as semantic errors, affecting the informativeness of their speech output. In contrast, people diagnosed with lvPPA presented with phonemic errors, reduced speech rate and impaired syntactical ability (Wilson et al., 2010). Dysfluencies such as false starts and sentence repairs were also reported to be typical in the connected speech of people with lvPPA, whilst incomplete sentences and increased use of pronouns are key features in both svPPA and lvPPA (e.g. Ash et al., 2013; Wilson et al., 2010). Reduced speech rate, phonemic and phonetic errors, as well as reduced syntactic complexity are key features in the connected speech of people with nfvPPA (Boschi et al., 2017). In contrast to PPA, AD is associated with global cognitive impairments, which then lead

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ISSN 1754-9507 print/ISSN 1754-9515 online
Published by Informa UK Limited, trading as Taylor & Francis Group
DOI: 10.1080/17549507.2018.1442498

to language dysfunction (Blair, Marczinski, Davis-Faroque, & Kertesz, 2007). Boschi et al. (2017) reported 36 studies that included people diagnosed with AD, mostly in the early stages of disease progression. Key deficits in the connected speech of people with AD included frequent hesitations and use of high-frequency words, ambiguous use of pronouns and lexical errors (e.g. Ahmed, Haigh, de Jager, & Garrard, 2013; Dijkstra et al., 2004). Although connected speech is recognised as a central component of language assessment with individuals with PPA and AD, it is not frequently used in clinical practice for these populations. Traditional language tasks used for individuals with PPA and AD are frequently based on single word or sentence processing (Boschi et al., 2017). This lack of translation into clinical practice is likely to reflect a multitude of challenges faced by speech-language pathologists.

Stability of connected speech in repeated samples

One key challenge is the uncertainty around whether connected speech is consistent over successive sampling, such that a single sample can be considered an accurate representation of an individual's language production capability. Knowing whether or not a measure, within a particular task, is likely to be representative for a person, such that a measure taken on one occasion is likely to be similar to that taken on another occasion within the assessment period window, is fundamental to the question of what, where and how often to sample connected speech in order to obtain a representative sample. Without repeated sampling, uncertainty as to the reproducibility of spoken production remains.

Only a small number of studies have explored the issue of stability of performance in repeated sampling of connected speech in neurodegenerative diseases. Stability is defined here as an absence of difference and/or variability of a linguistic feature in the same speaker, during repeated sampling, within a defined, and usually brief, period of time. Stable performance, demonstrated by a significant correlation between scores over successive samples and indicating an absence of any significant variation, would suggest that a score taken of that particular behaviour is representative for that individual. Within the literature, the term stability has been used consistently to denote this concept (e.g. Boyle, 2014; Ciccone, 2003; Fassbender, 2016; Hird, Brown, & Kirsner, 2006). In the PPA literature, one study has looked at the stability of specific discourse elements (Hird et al., 2006), while no studies have addressed stability with people with AD. Hird et al. (2006) investigated stability in repeated sampling of connected speech in PPA over three sessions, within a period of 2 months, by examining intra-individual confidence interval difference scores. A picture description, personal narrative and procedural narrative were used to sample connected speech; however, notably, one task was completed in each session, with no repetition of the different genres across sessions. The time between sessions, testing location and assessor information was not specified. Analysis of discourse samples included number of nouns, type token ratio (TTR), speech errors, pause analysis and efficiency of output. Efficiency was measured by correct information units per second (CIUs/sec), an adaptation of Nicholas and Brookshire's (1993) CIUs per minute (CIUs/min) analyses. Hird and colleagues reported stable performance across all measures, with the exception of communication efficiency, for five of the six people with PPA. The number of CIUs/sec showed a significant reduction across the three sessions, which the authors attributed to false starts and off task comments (i.e. participant comments focused on task performance). The authors suggested that this finding was indicative of general cognitive deficits in the early stages of PPA rather than a specific lexical processing deficit.

While one study of people with PPA has explored repeated sampling of connected speech, and no studies of people with AD, the post-stroke aphasia literature provides greater insight into this issue. Ciccone (2003) analysed the stability of connected speech samples in eight people with aphasia, comparing this to 10 healthy control participants. Four connected speech tasks including: picture description, picture sequence description (e.g. comic strips), personal narrative (e.g. "what did you do yesterday?"), and procedural narrative (e.g. "tell me how you would go about washing the dishes by hand") were repeated across eight testing sessions (approximately two sessions per week for a period of a month). To establish stability across the measures in connected speech, the SD and coefficient of variation were determined for communicative fluency and efficiency through number of words per second and CIUs/sec, respectively. Connected speech tasks were examined separately across each time point. In comparison to healthy control participants, people with aphasia, at a group level, showed greater overall variability in the number of words per second and CIUs/sec across the four connected speech tasks. Ciccone (2003) proposed that variability in communicative fluency and efficiency reflects the complex nature of connected speech which, through the involvement of a range of cognitive processes, would support the existence of multiple sources of variability. With some tasks requiring more cognitive resources than others, this would further account for variable performance between connected speech tasks. In contrast, Boyle (2014) reported stability in repeated sampling for 11 people with post-stroke aphasia, across three testing sessions, ranging from 2 to 7 days apart. Boyle (2014) used similar tasks to Ciccone (2003),

including stimuli from Brookshire and Nicholas (1994). The tasks described were two picture descriptions, one picture sequence, one personal narrative and one procedure, all of which were repeated in each session (Boyle, 2014). Examining, at a group level, correlation coefficients and the minimal detectable change score, stability was found for number of words, CIU informativeness (%CIUs), word finding difficulties and lexical diversity. Neither Boyle (2014) nor Ciccone (2003) examined individual differences of the participants. Cameron, Wambaugh, and Mauszycki (2010) collected connected speech samples, using 10 stimuli from Nicholas and Brookshire's materials (1993), for 11 people with post-stroke aphasia, also with a view to exploring stability over repeated sampling. All stimuli were repeated across five sessions (approximately 1 week apart, ranging from 1 to 42 days). Cameron and colleagues reported, at the group level, no significant differences between the means of all measures over time, using ANOVAs rather than correlations, including %CIUs and CIUs/min, mean number of words and number of words per minute (words/min), but stressed that greater variability across repeated sampling was found than had been in the original study by Nicholas and Brookshire (1993). Fassbender (2016) also used stimuli from Nicholas and Brookshire (1993), specifically two picture descriptions, one picture sequence, one personal narrative and one procedure, to investigate connected speech stability in 18 people with chronic post-stroke aphasia, looking at word class counts (nouns and verbs) and their relativity to each other, and %CIUs and CIUs/min. All elicitation tasks were repeated on three occasions: initial collection, 1 week later, and 4 weeks later. Using correlational statistics, for word class counts, stability across nouns was found for three participants and across verbs for four participants, with the relative diversity of lexical use also remaining stable. Other participants showed variability on word measures. Eight participants showed stability in CIUs/min, whereas five participants showed stability in %CIUs (Fassbender, 2016). Variability in repeated sampling for CIU measures was reported for the remaining participants, with the majority showing stability across two consecutive sessions but not for the third.

Connected speech tasks and genres

Integral to understanding the stability of connected speech is an understanding of the nature of the tasks used to elicit the speech sample and how these may impact performance. Boschi and colleagues, in addition to summarising the most frequently reported linguistic features of neurodegenerative diseases, reviewed the variety of tasks used to elicit connected speech and the plethora of linguistic measures used in analysis. Despite the identification of hallmark features, Boschi et al. (2017) reported that ambiguity was apparent in interpretation of behaviours, not least because a range of elicitation tasks and heterogeneous measures have been used to characterise the same linguistic features. They also identified the most commonly reported tasks for eliciting connected speech to be picture description, story narrative and various forms of conversation/interview, each of which bring different requirements. These different connected speech tasks are influenced by their unique purpose and inherently different internal structure of the genre elicited (Butt, Fahey, Spinks, & Yallop, 1999). Structured connected speech tasks usually limit speech output to one type of discourse, where topic, turn structure and type of response required is frequently predetermined. This type of connected speech is often referred to as semi-spontaneous speech, as a degree of structure is imposed and speech output is restricted to specific information (e.g. picture descriptions) or predefined stories (e.g. narrative production) (Prins & Bastiaanse, 2004). Spontaneous connected speech refers to everyday conversational exchanges between conversation partners (Boyle, 2011). Such dialogic contexts include tasks that impose less restriction on output with regard to time and information (Boschi et al., 2017). In comparison to semi-spontaneous tasks, conversational exchanges require more collaborative and interactive sampling where responsibility is shared, and communication partners may facilitate or hinder the exchange (Boyle, 2011).

Few previous studies have examined the connected speech performance of people diagnosed with PPA and AD across various elicitation tasks or discourse genres. Of the studies reviewed by Boschi and colleagues, only three studies investigated direct comparisons of connected speech tasks, indicative of limited cross-genre comparisons reported in the literature. Sajjadi, Patterson, Tomek, and Nestor (2012) compared the discourse performance in a semi-spontaneous interview and picture description for 16 people diagnosed with svPPA and 20 people diagnosed with AD. Semi-structured interviews were reported to be more sensitive in identifying morphology and syntax impairments, given the increased syntactic variability in semi-structured interviews, whilst picture descriptions were seen as more suitable for assessing semantic impairment, on the basis of increased noun usage in picture descriptions (Sajjadi et al., 2012). Ash et al. (2013) compared connected speech outcomes in a picture description and narrative task for people diagnosed with PPA (all variants) and behavioural variant frontotemporal dementia. In contrast to Sajjadi and colleagues, Ash et al. (2013) found that these tasks could be used interchangeably in identifying difficulties in semantic, phonetic and syntactic domains, as no significant differences were found between elicitation tasks. It may be possible to interpret the different findings

reported in these two studies as being associated with the varying degree of spontaneity and constraint associated with the tasks compared in each study. Specifically, Ash et al. (2013) compared two semi-spontaneous tasks, whereas Sajjadi et al. (2012) compared a semi-spontaneous to a spontaneous speech task. As category-specific impairments of lexical retrieval are common in svPPA (Harciarek & Kertesz, 2009), genre-specific differences may be expected to be dependent on the variations in restricted targets. Furthermore, Garrard and Forsyth (2010) proposed that spontaneous tasks (e.g. interviews) may impose less structure, allowing easier compensation for lexical difficulties in comparison to tasks such as picture descriptions which may lead to the production of simplified syntactic structures. Boschi et al. (2017) have called for further investigation comparing connected speech genres and tasks in different neurodegenerative conditions in order to optimise task and genre selection. Specifically, with the different genres highlighting different linguistic behaviours, it is also possible that the stability of language behaviours may change according to task and diagnostic group.

A greater understanding of language behaviours within the different genres and tasks used to sample connected speech will enable us to increase our confidence in using connected speech diagnostically to profile communication patterns in neurodegenerative disease and monitor change following intervention. This study investigated connected speech in PPA and AD, over a 3-week period, addressing the following questions:

(1) How stable is connected speech over three consecutive weeks in people diagnosed with PPA and AD on measures of lexical content, fluency and communicative informativeness and efficiency, both at a group and individual level?
(2) Are there differences in stability of these measures in different discourse sampling tasks, specifically everyday monologues, narrative and picture description, over this time period?

Method

Participants

Nine adults, aged between 55 and 86 years ($M = 64.7$), were recruited via private neurologists, geriatricians and speech-language pathologists working in Perth, Australia. Three participants were diagnosed with svPPA, three participants with lvPPA, and three participants with AD featuring prominent language difficulties. Participants were diagnosed, according to Mesulam's (2001) criteria, by their treating neurologist or geriatrician through a combination of brain imaging and cognitive assessment. Ethics approval was received from the Curtin University Human Research Ethics Committee (HR218/2015). All participants were in the pre-clinical or clinical stages of disease progression, as determined by a mild to moderate score on the Addenbrooke's Cognitive Examination – third edition (ACE-III; Mioshi, Dawson, Mitchell, Arnold, & Hodges, 2006). Participants were native English-speaking, had no reported hearing impairments, and were not receiving speech-language therapy during the study period. Participants enrolled in medication-based studies were not excluded where dosage had been stable for a minimum of 3 months.

Participant profile

All participants were tested on a language and cognitive battery. The assessment battery comprised the ACE-III (Mioshi et al., 2006), Pyramids and Palm Trees Test (PPTT; Howard & Patterson, 1992), Kissing and Dancing Test (K&DT; Bak & Hodges, 2003) and Northwestern Naming Battery (NNB; Thompson & Weintraub, 2014) (see Table I for psychometric data, assessment results and demographic information).

SvPPA participants

Participant 1 (P1) was a 62-year-old man who presented with a 5-year history of word finding difficulties and more recent comprehension difficulties. At the time of the study, he was still working as a metallurgist. Participant 2 (P2) was a 55-year-old man with a 4-year history of word finding and comprehension difficulty. Due to increasing communication concerns, he had retired from his engineering position 2 years prior to the study. Participant 3 (P3) was a 72-year-old man who reported difficulty finding words and retaining newly acquired words since the age of 60. He had retired from his position as a director of children's homes.

Based on the ACE-III results, P1's overall score (84/100) was just above the cut-off score, suggestive of relatively intact cognition (see Table I). In contrast, P3 scored 34/100, with impairment across all sections, particularly fluency (1/14) and language (4/26). Due to the impact of his semantic deficit, P2 was unable to complete the ACE-III. On the language assessment, P1 performed just below the normative cut-off on the PPTT and within normal limits on the K&DT. Both P2 and P3 presented with impaired performance on noun and verb semantic conceptual knowledge, however, both performed slighter poorer for nouns (see Table I). Impaired confrontation naming was found for all participants, however, more severe for participants 2 and 3. P1 achieved ceiling level on auditory comprehension, whereas P2 and P3 performed below normal limits. Ceiling performance was achieved by all three participants on the non-word and word repetition tasks.

LvPPA participants

Participant 4 (P4) was a 60-year-old female who reported a 3-year history of increasing difficulty

Table I. Demographic and background cognitive and language information for participants.

	P1	P2	P3	P4	P5	P6	P7	P8	P9	Normative data *M* (*SD*)
Age (years)	62	55	72	60	70	59	86	60	58	–
Gender	M	M	M	F	M	F	M	F	M	–
Diagnosis	svPPA	svPPA	svPPA	lvPPA	lvPPA	lvPPA	AD	AD	AD	–
Education (years)	18	16	10	16	14	9	9	13	16	–
Time post diagnosis (months)	44	51	50	38	38	12	5	15	58	–
Cognitive screen[a] (/100)	84	–	34	76	59	42	62	47	23	58.7 (21.4)[b]
Language measures										
Conceptual semantics										
Nouns[c] (/52)	44	29	33	52	50	52	45	44	32	47.6 (2.6)[d]
Verbs[e] (/52)	49	42	45	51	48	51	45	43	34	48.3 (3.2)[d]
Confrontation naming[f]										
Nouns (/16)	11	0	0	16	15	16	15	16	15	12.1(5.3)[g]
Verbs (/16)	14	3	6	15	12	13	13	16	11	12.6 (3.9)[g]
Auditory comprehension[f]										
Nouns (/12)	12	1	5	12	12	12	12	12	11	11.2 (1.9)[g]
Verbs (/12)	12	6	9	12	12	12	12	12	12	11.5 (1.1)[g]
Repetition[f]										
Word (/21)	21	21	21	21	21	21	21	21	21	20.6 (1.0)[g]
Non-word (/10)	10	10	10	8	8	10	10	10	10	8.6 (1.9)[g]

[a]Addenbrooke's Cognitive 422 Examination – third edition (ACE-III).
[b]Normative data for PPA (Hsieh, Schubert, Hoon, Mioshi, & Hodges, 2013). Cut-off at overall score 82 indicates 100:1 likelihood of dementia (Mioshi, Dawson, Mitchell, Arnold, & Hodges, 2006).
[c]Pyramids and Palm Trees Test (PPTT).
[d]Normative data based on healthy sample aged 20–63 years (Mansur, Carthery-Goulart, Bahia, Bak, & Nitrini, 2013).
[e]Kissing and Dancing Test (K&DT).
[f]Northwestern Naming Battery (NNB).
[g]Adapted from the NNB normative data for 37 individuals with PPA aged 48–81 years (Thompson & Weintraub, 2014).

pronouncing words as well as occasional word finding difficulties. Due to increasing language difficulties, she retired from her position as deputy principal 2 years prior to the study. Participant 5 (P5) was a 70-year-old ex-army man who presented with a 4-year history of impaired word finding and pronunciation. Similarly, participant 6 (P6), a 59-year-old female, presented with a 2-year history of word finding and pronunciation difficulties. She also reported occasional comprehension difficulties, and had retired from her work in customer service.

Performance on the ACE-III indicated variable performance in cognitive functioning across participants 4–6 (see Table I). P5 presented with difficulties across all sections, whereas P4 and P6 presented with most prominent impairment in the memory and language sections. Semantic conceptual knowledge was intact for nouns and verbs for all three participants based on the PPTT and the K&DT. Confrontation naming of nouns was intact for all participants, however, participants 5 and 6 presented with mild verb naming deficits. Mild difficulties were found on the non-word repetition task for participants 4 and 5, whilst performing at ceiling level for word repetition.

AD participants

Participant 7 (P7), a retired bank teller, was an 86-year-old man who reported a 4-year history of short-term memory and marked word finding difficulties despite intact comprehension. Participant 8 (P8) was a 60-year-old female who presented with a 2-year history of impaired comprehension and short-term memory difficulties. Due to increasing communication difficulties, she had retired from her work as a garden and nursery worker, and had also withdrawn from various volunteering positions. Similarly, participant 9 (P9) was a 58-year-old man who presented with a 6-year history of impaired comprehension and short-term memory. He further demonstrated impaired visuospatial processing and word finding difficulties, and had retired from his position as a geologist.

Participants 7, 8 and 9 all showed impaired cognition across all sections of the ACE-III, in particular memory and fluency (see Table I). Performance on the PPTT and K&DT indicated mild impairment in semantic conceptual knowledge for P7 and P8, whereas P9 showed moderate impairment for both nouns and verbs. P8 demonstrated ceiling performance on confrontation naming, whereas P7 and P9 showed mild verb naming impairments. Auditory comprehension, non-word and word repetition was intact for all three participants.

Procedure

Design

A repeated measure, within groups, research design was used to investigate the stability of connected speech measures. Connected speech sampling was repeated on three occasions, on average 7 days apart (ranging from 4 to 8 days), with no intervening speech-language therapy. Cognitive and language testing was completed over two sessions within this 3-week period.

Assessment

Three occasions of connected speech samples were collected from each participant. Connected speech sampling consisted of: (a) nine everyday monologues from the Curtin University Discourse Protocol (CUDP) (Whitworth, Claessen, Leitão, & Webster, 2015) including recounts (weekend, Christmas, past injury), procedures (changing a lightbulb, planning a meal, scrambling eggs) and expositions (global warming, obesity, bullying) (one topic from each genre elicited on each occasion across the three sessions); (b) a narrative monologue (Cinderella); and (c) two picture description tasks; Cookie Theft picture (Goodglass & Kaplan, 1983) and Birthday Cake picture (Nicholas & Brookshire, 1993). All monologues were repeated on all three occasions and were elicited in accordance with the protocol guidelines of the CUDP. Assessors were independent, qualified speech-language pathologists blinded to the participant's diagnosis. All samples were audio recorded using an iPad, in each participant's home. Dialogue between tasks were kept to a minimum. Connected speech samples were transcribed by the first author and entered into Systematic Analysis of Language Transcripts computer software for analysis (Miller & Iglesias, 2008). All samples were coded for: (1) lexical content, including TTR, number of different words (NDW), pronouns, nouns, light verbs and heavy verbs; (2) fluency, including %mazes (i.e. filled pauses) and words/min; and (3) communicative informativeness and efficiency, including %CIUs and CIUs/min (Nicholas & Brookshire, 1993). Words/min, TTR, and NDW were not included for genre analysis. To ensure a representative overview of lexical content, light and heavy verbs were considered on the basis of their varying semantic component. For example, heavy verbs with richer semantic specificity (e.g. dance, cook, drink) were compared to light verbs (e.g. put, do, get) (Barde, Schwartz, & Boronat, 2006). Inter-rater reliability of greater than 80% for coding was established for 20% of the connected speech samples by three trained clinicians.

Data analysis

Test–retest stability of connected speech measures across the three consecutive samples was tested across the conditions of everyday monologue (recount, procedure and exposition tasks collapsed), narrative and picture description. The intra-class correlation (ICC) is a measure of stability which captures both the consistency and agreement components (Liu, Tang, Chen, Feng, & Tu, 2016). According to Koo and Li (2016), the appropriate ICC for measuring stability should use a two-way mixed-effects model in conjunction with averaged measures and absolute agreement. In the current study, ICC estimates of stability and their 95% confident intervals were therefore calculated using SPSS (version 21) based on a two-way mixed-effects model, absolute-agreement and averaged measures ($k = 3$ assessments). Cicchetti (1994) gives the following guidelines for interpretation of the ICC values: less than 0.40 = poor, between 0.40 and 0.59 = fair, between 0.60 and 0.74 = good and between 0.75 and 1.00 = excellent. To determine stability for individual cases, Tau U analyses and Fisher's Exact Variance test with a Poisson Distribution were run.

Result

Raw data for individual participants for all connected speech measures is presented in the Supplementary material (A, B and C).

Stability of connected speech at the group level

Stability of lexical content. Stability of lexical content in connected speech was measured using TTR, NDW and word class counts of pronouns, nouns, light verbs and heavy verbs. Across the three assessments, excellent stability was found for all measures, except nouns within picture descriptions, across the three discourse genres (ICC = 0.784 to 0.962, $p = 0.005$ to <0.001) (see Table II). Nouns within picture descriptions showed good stability although this was not significant (ICC = 0.614, $p = 0.051$).

Stability of fluency. Stability of fluency in connected speech was measured using %mazes and words/min. An excellent degree of stability was

Table II. Intra-class correlation (ICC), confidence intervals and significance values for lexical content measures.

		95% confidence interval		
Lexical content measures (average measures)	ICC	Lower bound	Upper bound	*p*
Type token ratio				
Everyday monologue	0.907	0.714	0.977	0.000
Narrative	0.801	0.372	0.951	0.004
Picture description	0.784	0.345	0.947	0.005
No. of different words				
Everyday monologue	0.820	0.473	0.955	0.001
Narrative	0.982	0.943	0.996	0.000
Picture description	0.810	0.424	0.953	0.003
Pronouns				
Everyday monologue	0.839	0.509	0.960	0.001
Narrative	0.972	0.913	0.993	0.000
Picture description	0.934	0.797	0.984	0.000
Nouns				
Everyday monologue	0.903	0.710	0.976	0.000
Narrative	0.988	0.963	0.997	0.000
Picture description	0.614	0.175	0.905	0.051
Light verbs				
Everyday monologue	0.794	0.386	0.949	0.003
Narrative	0.974	0.918	0.994	0.000
Picture description	0.872	0.587	0.969	0.000
Heavy verbs				
Everyday monologue	0.869	0.601	0.968	0.000
Narrative	0.989	0.968	0.997	0.000
Picture description	0.962	0.881	0.991	0.000

<0.5 is indicative of poor reliability; 0.5–0.75 indicates moderate reliability; 0.75–0.9 indicates good reliability; >0.90 indicates excellent reliability (Cicchetti, 1994).

Table III. Intra-class correlation (ICC), confidence intervals and significance values for fluency measures.

Fluency measures (average measures)	ICC	95% confidence interval Lower bound	Upper bound	*p*
% Mazes				
Everyday monologue	0.978	0.933	0.995	0.000
Narrative	0.619	0.316	0.910	0.060
Picture description	0.972	0.915	0.993	0.000
Words/min				
Everyday monologue	0.902	0.706	0.976	0.000
Narrative	0.973	0.919	0.993	0.000
Picture description	0.973	0.919	0.993	0.000

<0.5 is indicative of poor reliability; 0.5–0.75 indicates moderate reliability; 0.75–0.9 indicates good reliability; >0.90 indicates excellent reliability (Cicchetti, 1994).

Table IV. Intra-class correlation (ICC), confidence intervals and significance values for communicative informativeness and efficiency measures.

Communicative informativeness and efficiency measures (average measures)	ICC	95% confidence interval Lower bound	Upper bound	*p*
%CIUs				
Everyday monologue	0.678	0.012	0.921	0.027
Narrative	0.678	0.012	0.912	0.027
Picture description	0.874	0.605	0.969	0.000
CIUs/min				
Everyday monologue	0.760	0.267	0.941	0.008
Narrative	0.984	0.952	0.996	0.000
Picture description	0.884	0.638	0.971	0.000

<0.5 is indicative of poor reliability; 0.5–0.75 indicates moderate reliability; 0.75–0.9 indicates good reliability; >0.90 indicates excellent reliability (Cicchetti, 1994).

found across the three time points within everyday monologue, %mazes (ICC = 0.978, $p < 0.001$) and words/min (ICC = 0.902, $p < 0.001$) and picture description, %mazes (ICC = 0.972, $p < 0.001$) and words/min (ICC = 0.973, $p < 0.001$) (see Table III).

Stability of communicative informativeness and efficiency. Stability of communicative informativeness and effiency in connected speech was measured using %CIUs and CIUs/min, respectively. Across the three assessments, excellent stability was found for %CIUs within picture description only (ICC = 0.874, $p < 0.001$), whilst everyday monologue and narrative showed good stability (ICC = 0.678, $p < 0.027$ for both genres) (see Table IV). Excellent stability was found for CIUs/min within everyday monologue (ICC = 0.760, $p = 0.008$), narrative (ICC = 0.984, $p < 0.001$) and picture description (ICC = 0.884, $p < 0.001$).

Stability of connected speech at an individual level

Where available, participants' data were analysed individually using Tau U (Vannest, Parker & Gonen, 2011), to measure the strength and direction of association of time and connected speech measures for picture description, everyday monologues and narrative. Narrative analyses were not completed for P2, P3 and P6 in which narrative samples were unable to be elicited, as well as P5 and P9 in which narrative samples were inconsistently elicited. For picture description and everyday monologue, no significant differences were found for all participants over the three samples (see Supplementary material D). For narrative, no statistically significant differences were found for the four participants who completed three consistent samples. For P5 and P9, the Fisher's Exact Variance Test with a Poisson Distribution was used to determine significant differences over two time points. No significant differences in the connected speech measures were found (see Supplementary material E), except for CIUs/min for P5 ($p = 0.036$).

Discussion

This study investigated the stability of connected speech over three consecutive weeks in people diagnosed with PPA and AD on measures of lexical content, fluency and communicative informativeness and efficiency. Accurate and reliable measurement of the characteristics of connected speech is essential if discourse is to be used in research and clinical practice, but must be coupled with knowledge that a sample is representative of a person's discourse. For individuals with PPA and AD, understanding whether stability can be observed within a relatively short period is particularly important in the context of both the profile of the disorder and its progressive nature.

Stability of connected speech measures

For lexical content, TTR, NDW, pronouns, nouns, light verbs and heavy verbs all showed excellent stability. For nouns, good stability was found for one of the discourse genres (i.e. picture description); however, this was not significant. Group-level analyses were supported by individual-level analyses, in which no significant evidence of variability was found for lexical content measures; this was across all genres. For fluency measures, both %mazes and words/min showed excellent stability across the three assessments for at least two genre types. Similar to the lexical content measures, fluency group-level analyses were supported by individual-level analyses, with no significant results that indicated variability. For communicative efficiency, excellent stability was consistently found, whilst informativeness varied across the different genres, showing levels of stability that ranged from good to excellent. Again, group-level analyses were supported by individual analyses with one exception. For P5, CIUs/min was found to be significant over two time points (where only two samples could be obtained).

Findings are consistent with reports of stability in the post-stroke literature, despite hypotheses that stability may be reduced in progressive populations due to cognitive factors. Specifically, findings are consistent with Boyle (2014) who reported stable %CIUs and lexical diversity, as well as Cameron et al. (2010) who found stable CIUs/min, %CIUs and words/min, in adults with post-stroke aphasia. The results conflict, however, with Hird et al. (2006) who reported significant variability of CIUs/sec for participants with PPA. It is important to note that Hird and colleagues did not repeat discourse genres, rather, sampled different genres across each of the three sessions. In contrast, Boyle (2014) and Cameron et al. (2010) repeated the same genres and tasks across sessions, similar to the current study. Assessing a range of genres over sessions has clear implications for performance, as Ciccone (2003) proposed that different genres may require more cognitive resources than others, resulting in differential performance. This explanation of variability differs to that proposed by Hird and colleagues, who specifically proposed that poor stability of communication efficiency could be explained by general cognitive deficits.

Stability across genres

Everyday monologues showed excellent stability for all lexical content measures, fluency measures and communicative efficiency, whilst good stability was found for communicative informativeness. Similarly, within the narrative discourse genre, excellent stability was found for all lexical content measures, one fluency measure (words/min) and communicative efficiency. For the remaining measures, specifically %mazes and %CIUs, good stability was found within the narrative genre. Lastly, picture description showed excellent stability for most lexical content measures (with the exception of nouns), fluency measures and communicative informativeness and efficiency.

Of the tasks used to sample the different genres, providing a narrative was the only task which participants had difficulty completing, with five participants being partially or completely unable to produce a sample across the 3 weeks. An interesting finding was that two of the three participants who consistently did not produce narrative samples demonstrated the most significant semantic memory deficits. Specifically, P2 and P3, both diagnosed with svPPA, showed severe impairments on the K&DT, confrontation naming and auditory comprehension subtests of the NNB. This may highlight the impact of semantic memory deficits on the ability to produce connected speech for tasks, such as narrative, that impose highly specific semantic information. The comprehension of words and concepts may have been impaired to the extent that capacity to both follow instructions and access vocabulary in order to complete the task was highly compromised. This suggests that clinicians and researchers should exercise caution in using narrative in isolation when assessing connected speech.

Stability across individual participants

This study also examined the stability of each of the nine participants at an individual level and found all participants were stable over the 3-week period across genres and measures. While some individuals were unable to complete the narrative task, those participants that could complete either two or three samples were stable on all measures. One exception was seen in the narrative samples provided by one participant (P5). Participant 5 demonstrated significant changes in communicative efficiency (CIUs/min) between his two narrative samples. Notably, there were no differences seen in stability across the two diagnostic groups (PPA and AD) in this current study. While sample sizes were small, these findings suggest that stability can be seen irrespective of PPA or AD profiles. The current study reveals the importance of evaluating stability at both group and individual levels.

Clinical implications

The current study provides preliminary evidence to suggest that a single baseline connected speech sample may be sufficient in providing accurate baseline data, depending on the connected speech measures and genres that are to be analysed from the sample. Single baseline collection is appealing clinically for a number of reasons. It has the potential to increase the efficiency of data collection during assessment whilst maintaining confidence in the reliability of the data obtained. The less time consuming nature of a single connected speech sample has implications for both the clinician who can gain valuable diagnostic information, and the client who may find connected speech sampling less taxing than formalised measures of assessment. The findings are consistent with previous reports of stability from the post-stroke literature but contrast with the findings of one study with adults who had PPA. The current study provides evidence of stability on language and communicative measures in genres used to routinely sample connected speech in clinical settings.

Recommendations for future research

From this study, the need for a greater understanding of the connected speech genres and tasks used in clinical practice, as well as of the linguistic measures themselves, is evident. The findings of this study highlight the stability of connected speech measures in adults with PPA and AD. Future research may explore the influence of

connected speech genre and task selection on linguistic measures. In addition, this study highlights the need to further develop our understanding of the internal structure of discourse genres. Sajjadi et al. (2012) proposed, for example, that interviews may be more useful in identifying syntactic difficulties while picture descriptions may reveal semantic difficulties. Further research is required to determine how discourse genres and the specific tasks vary and ultimately influence connected speech production. This understanding will likely guide task and discourse genre selection, with specific consideration given to both the connected speech measures used and the purpose of language sampling (e.g. diagnostic profiling vs. establishing baseline performance). In addition to further consideration of the impact of connected speech genre, more research is also required to explore the stability and suitability of language measures in individuals with PPA and AD. Exploring the stability of other language measures (e.g. grammatical structures) may enhance current understanding about the language stability, and variability, in adults with PPA and AD.

Limitations

While this study has enabled us to look at a small number of participants in considerable depth, the sample size limits external validity of the study. While the rare nature of PPA does restrict recruitment, replication is recommended with a larger sample size. Analysis of psychometric data on the CUPD with healthy speakers is not currently available.

Conclusion

This is the first study to explore the stability of a range of discourse genres in people with PPA and AD with a view to exploring the clinical utility of connected speech evaluation in the assessment process. Findings of excellent levels of stability across a range of measures, taken on three occasions over a 3-week period, provide guidance to clinicians as to the stability of language use by adults with PPA and AD. While further investigation across a larger sample is indicated, these findings provide early support for a single collection point in time potentially being sufficient to provide representative baseline data for individuals from these populations, dependent on both measure and genres that are to be analysed from the sample.

Acknowledgements

We would like to thank all of the participants who took part in this research. We would also like to thank speech-language pathology students, Daphlyn Goh and Sallina Le, for their work on this project.

Declaration of interest

The authors report no declarations of interest.

This work was supported by the Dementia Australia Research Foundation and Curtin University through the Australian Government Research Training Program.

Supplementary material

Supplemental data for this article can be accessed at https://doi.org/10.1080/17549507.2018.1442498.

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PHASE THREE

Evaluation of a Self-Cueing Lexical Retrieval Intervention and Exploration of Intervention Experiences

Chapter 5

The findings of Phase One and Phase Two of the research program informed the intervention design that is evaluated across two studies in Phase Three. Chapter 5, comprising one study which is submitted and under review and therefore presented in chapter form, presents the findings of the first study in Phase Three of the research that aimed to evaluate the implementation of a metacognitive strategy intervention for lexical retrieval, specifically, a strategic self-cueing approach with embedded nonlinguistic cognitive scaffolds.

Study Overview

While the growing body of treatment evidence is testimony to the transferability of rehabilitative-style lexical retrieval interventions to neurodegenerative conditions, Phase One revealed that the diversity of approaches applied and the associated mechanisms of change proposed within these do not allow a clear understanding of which approaches are most effective, whether these are effective for different reasons than when used in the post-stroke aphasia population, and whether generalisation patterns are similar. Although there have been a limited number of studies conducted, promise for enhanced generalisation gains following self-cueing approaches was reported in Phase One. This approach has been utilised in the PPA literature in which three case series studies have reported evidence of within level effects, specifically, improved naming of untreated items, following a strategic self-cueing approach (Beales et al., 2016; Beeson et al., 2011; Henry et al., 2013). Moreover, Henry et al. (2019) conducted a group level study (nine svPPA and nine lvPPA participants), which trained a self-cueing strategy via systematic retrieval of linguistic knowledge (semantic, phonological, and orthographic information), as well as one cognitive-cue (episodic/autobiographical information), to promote lexical retrieval. Henry et al. (2019) reported improved naming of untreated items for the combined PPA cohort and attributed outcomes to the strategic nature of intervention. Given the promising generalisation findings for strategy-based interventions with an underlying cognitive-relay mechanism reported in Phase One, this study investigates the outcomes, at both the diagnostic group and individual level, of a metacognitive strategic self-cueing intervention for people with PPA and AD. Informed by the findings of Phase Two of this research program, consideration of working memory, as well as other cognitive functions, were addressed through incorporation of

individualised cognitive scaffolds. Additionally, to address the need for understanding of discursive generalisation patterns (Cadório et al., 2017), evaluation of across level generalisation was examined through connected speech measures, as supported by evidence of sampling stability reported in Phase Three.

Maximising Access Across Word Classes and Connected Speech

While most lexical retrieval interventions have focused on nouns, a large portion of words used frequently in daily communication include those from other word classes, such as adjectives, adverbs, verbs, and pronouns (Renvall et al., 2013), and have been increasingly introduced into treatment protocols. The inclusion of multiple word classes as therapy targets supports the individual to express a wider range of communicative intentions while also having the potential to facilitate argument and sentence structure through their syntactic relationships. Two PPA studies have reported explicit inclusion of adjectives in the treatment stimuli (Beales et al., 2016; McNeil et al., 1995). Both studies reported improved naming of untreated adjectives, however, this was not supported by statistical analysis in the McNeil et al. (1995) study. Following a strategic self-cueing approach, Beales et al. (2016) reported patterns of within level generalisation outcomes for nouns, verbs, and adjectives, however, importantly, statistically significant gains were variable across both the individual participants and word classes. These studies have indicated that a range of word classes can be responsive to intervention, and a degree of within level generalisation can occur, but that the strength and pattern of transfer remains variable.

Beales et al. (2016) also found different across level generalisation patterns for participants. The three participants diagnosed with svPPA demonstrated a significant increase in communicative informativeness and efficiency, in contrast to the participant with lvPPA who remained stable. Beales et al. (2016) proposed that the inconsistent patterns of across level generalisation may have been contributed to by the absence of intervention focus in connected speech contexts, calling for future research to extend beyond lexical retrieval in isolation and include multiple levels of language processing. For example, in a multilevel discourse intervention for two people with PPA (one lvPPA and one svPPA) (Whitworth et al., 2017), significant gains in the amount of overall connected speech output were reported, along with overall noun and verb usage, in novel everyday monologues for both participants.

Whitworth et al. (2017) suggested that targeting lexical retrieval within discourse contexts, with an explicit focus on sentence and discourse structure, may be critical to facilitate greater lexical retrieval in everyday connected speech. Notably, while this study aimed to improve discourse structure rather than simply provide opportunities for word retrieval in connected speech, it drew attention to the potential across level gain when systematically incorporated into therapy.

Nonlinguistic Cognition in Treatment

A notable conclusion from Phase One of this research was that limited exploration had taken place, to date, as to the role of nonlinguistic cognitive functions with respect to assessment, planning intervention, and interpreting intervention outcomes. Only autobiographical memory and episodic memory had been incorporated into PPA and AD lexical retrieval interventions, most commonly through the form of personally relevant treatment items (e.g. Clare et al., 1999), spaced retrieval principles (e.g. Bier et al., 2009), and episodic/autobiographical self-cues (e.g. Beales et al., 2016). While other nonlinguistic cognitive functions, such as attention, working memory, and visual recognition memory, are yet to be explored within the PPA and AD intervention literature, they are likely to impact an individual's ability to engage in therapy, as well as their capacity to take on strategies. For example, for individuals with post-stroke aphasia, Fillingham et al. (2006) found greater improvement following intervention for those participants who had less impaired working memory and executive function abilities prior to intervention, but not necessarily participants with greater language abilities. Executive functions have explicitly been found to predict an individual's ability to learn strategies and apply these in contexts outside of intervention (Nicholas et al., 2005). Moreover, visuospatial working memory has been reported to be highly predictive of an individual's response to lexical retrieval treatment (Harnish & Lundine, 2015).

With cognition often playing a significant role in progressive conditions, awareness of potential cognitive deficits and the demands of intervention on cognitive processes would seem to be paramount with these clinical populations. Attention is reported to be one of the early nonlinguistic cognitive functions affected in AD, following the initial amnestic stage (Perry & Hodges, 1999). Deficits in nonlinguistic cognitive functions have been shown to emerge primarily in the

intermediate and advanced stages of PPA (Bettcher & Sturm, 2014) and differ across the variants of the condition. Verbal working memory deficits have been reported in lvPPA (Bettcher & Sturm, 2014), in comparison to largely intact performance in svPPA as found in Phase Two. Furthermore, mild deficits in visuospatial processing have been reported in lvPPA, while visual memory is typically intact in comparison to verbal episodic memory (Bettcher & Sturm, 2014). Bettcher and Sturm (2014) report sparing of verbal episodic memory in svPPA, despite performance on tasks frequently being reduced due to impoverished semantic knowledge. Although visual memory is typically preserved in the early stages of svPPA, deficits are suggested to arise at later stages due to impaired visual recognition. Consequently, establishing an individual's strategic competence would seem to be critical for tailored treatment approaches which enhance generalisation outcomes via strategy use (Canale & Swain, 1980).

Communication Partners in Treatment

In a recent systematic review of functional communication interventions for PPA, one of the key intervention components identified for prioritisation was strategy practice with a communication partner (Volkmer et al., 2019). The importance of the use of strategy and natural aspects of communication into intervention has been well documented in the post-stroke aphasia literature (e.g. Green, 1982). Green (1982) proposed that the participant's desire to communicate is fundamental to generalisation of strategy use, which is optimised through the involvement of communication partners and connected speech contexts. In the PPA literature, Grasso et al. (2017) reported a lexical retrieval intervention, involving both a clinician- and caregiver-administered treatment phase, with one individual diagnosed with mild cognitive impairment and one diagnosed with lvPPA. Interestingly, no significant differences in treatment effects were found between the clinician-administered phase and the caregiver-administered phase, with significant improvements in naming treated items observed following both conditions. Involvement of communication partners has also been explored in the AD literature through studies that have specifically targeted language stimulation programs administered through caregiving dyads (e.g. Quayhagen & Quayhagen, 2001). Results from these dyadic interventions have shown improvement in language of the person with AD, supporting the role of spousal caregivers as active ingredients in

cognitive-linguistic remediation in dementia syndromes. Practice involving a communication partner therefore not only has the potential to increase opportunities for strategy use in the home environment and other functional contexts, thereby increasing dose, but may maximise salience of topics and practice of functional words, a critical principle of relearning and neuroplasticity (Maas et al., 2008). In this regard, the roles that partners can play in facilitating generalisation are many-fold and as such, including partners in therapy protocols would seem to be advantageous.

Aims

An intervention protocol was used that built upon key elements of an earlier self-cueing intervention study (Beales et al., 2016) and combined these with critical new elements. Intervention ingredients carried forward included (1) a strategic self-cueing approach, (2) repeated practice of the strategy in retrieving treated items to reinforce skill acquisition (Maas et al., 2008), (3) strengthening of lexical-semantic, lexical-syntactic, phonological, orthographical, and autobiographical associations of treated items (Beales et al., 2016), with autobiographical associations capitalising on self-reference and maximising salience, and (4) inclusion of nouns, verbs, and adjectives to support access to a wider range of communicative intentions, and maximise facilitation of syntactic and argument structure.

While within and across level generalisation were seen following the original protocol, this was inconsistent across word classes, across the language levels, and across participants. To enhance treatment and generalisation outcomes, we expanded the intervention protocol in a number of ways. First, to address the limited exploration to date of the role of nonlinguistic cognitive functions with respect to planning intervention, individualised cognitive scaffolding of executive function, visual attention, working memory, verbal episodic memory, and visual recognition memory, based on participants' pre-intervention neuropsychological assessment, was incorporated to facilitate strategy use. Second, the inclusion of communication partners into the therapy sessions, complemented by home practice, aimed to promote generalised strategy use within everyday contexts, maximise salience of topics and novel words selected to practice the strategies, and increase therapy dose. Finally, the use of the strategy approach in dyadic interaction with communication partners promoted opportunities to use self-cueing strategies within a connected

speech context. Given that people with AD are more likely to present with impaired nonlinguistic cognitive functions, we anticipated that people with PPA were more likely to have increased capacity for strategic competence and therefore show greater gains following a strategy use intervention, despite individualised cognitive scaffolds introduced to compensate for diagnostic variation across the groups.

We hypothesised that all diagnostic groups would demonstrate a significant increase in retrieval of treated items in each of the three word classes but with gains for participants with PPA likely to be higher than those with AD. Due to the inclusion of additional components to enhance generalisation, we further hypothesised that both within level and across level generalisation would be seen for all diagnostic groups, demonstrated in a significant increase in retrieval of untreated items across all word classes, and a significant increase in communicative informativeness and efficiency in connected speech. Similarly to diagnostic group hypotheses, we predicated that on individual analysis all participants would demonstrate an increase in retrieval of treated and untreated words, however, based on the findings of Beales et al. (2016), variability was anticipated but generalised gains expected to be more frequent than in the earlier study. Finally, we expected nonlinguistic cognitive performance to remain stable pre- and post-intervention for all diagnostic groups as cognitive functions were not directly targeted.

Method

Design and Participants

A multiple baseline across-behaviors design was used in which participants acted as their own control. All aspects of the Template for Intervention Description and Replication (TIDieR) were addressed (Hoffmann et al., 2014). Strategies to improve fidelity were implemented through use of intervention parameters, clinician record sheet for intervention sessions, and a treatment adherence rating protocol. This study was approved by the Curtin University Human Research Ethics Committee (HR218/2015).

Twelve adults, aged between 55 and 86 years (M = 65.5), with a diagnosis of PPA or AD were recruited to the study via private neurologists and geriatricians from rural, regional, and metropolitan areas of Western Australia. Four participants had a diagnosis of svPPA, four lvPPA, and four AD. Individuals were required to have a diagnosis of PPA (all variants) or AD (amnestic presentation), as determined by a mild to moderate score on the Addenbrooke's Cognitive Examination – third

edition (ACE-III; Hsieh et al., 2013; So et al., 2018). Participants had no reported. hearing impairments, were native English-speaking, and were not receiving other speech-language pathology services during the study period.

Pre-intervention language assessments were completed to characterise the sample (see Table 2). Impaired noun and verb conceptual semantics was found for two participants diagnosed with svPPA (P2, P3) and two with AD (P11, P12), while mild deficits were found for two participants with AD (P9, P10). P1 (svPPA) also demonstrated a mild deficit with nouns but normal performance with verb conceptual semantics. All participants diagnosed with lvPPA and P4 (svPPA) performed within normal limits for conceptual noun and verb semantics. Impaired auditory comprehension for nouns and verbs was found for two participants with svPPA (P2, P3) and one with AD (P11). All other participants performed within normal limits. Two participants with svPPA who had shown conceptual deficits (P2, P3) showed marked impairment on noun and verb confrontation naming, while mild deficits for nouns only were seen for the other participants diagnosed with svPPA (P1, P4). Mild impairments were also found for participants diagnosed with lvPPA (P6, P7, P8) and AD (P9, P11, P12).

Table 2

Characterisation of PPA and AD participants according to demographic information and pre-intervention performance on linguistic assessments

	Participant												Normative data
	1	2	3	4	5	6	7	8	9	10	11	12	*M (SD)*
Age (years)	62	55	72	72	60	70	64	59	86	60	68	58	n/a
Gender	M	M	M	M	F	M	F	F	M	F	M	M	n/a
Diagnosis	svPPA	svPPA	svPPA	svPPA	lvPPA	lvPPA	lvPPA	lvPPA	AD	AD	AD	AD	n/a
Education (years)	18	16	10	14	16	14	10	9	9	13	12	16	n/a
Time post diagnosis (years)	2	3	3	1	3	3	1	1	1	2	4	4	n/a
Handedness	R	R	R	R	R	R	R	R	R	R	R	R	n/a
ACE - III (/100)	84	23	34	74	76	59	70	42	62	47	26	23	58.7(21.4)[1]
PPTT (/52)	44	29	33	48	52	50	49	52	45	44	26	32	47.6(2.6)[2]
K&DT (/52)	49	42	45	49	51	48	49	51	45	43	28	34	48.3(3.2)[2]
NNB													
Auditory comprehension (/12 nouns: /12 verbs)	12:12	1:6	5:9	12:12	12:12	12:12	12:12	12:12	12:12	12:12	9:8	11:12	11.2(1.9)[3] 11.5(1.1)[3]
Confrontation naming (/16 nouns: /16 verbs)	11:14	0:3	0:6	11:15	16:15	15:12	14:16	16:13	15:13	16:16	13:9	15:11	12.1(5.3)[3] 12.6(3.9)[3]

Note. ACE-III = Addenbrooke's Cognitive Examination third edition, PPTT = Pyramids and Palm Trees Test (Howard & Patterson, 1992), K&DT = Kissing and Dancing Test (Bak & Hodges, 2003), NNB = Northwestern Naming Battery (Thompson & Weintraub, 2014), NAVS = Northwestern Assessment of Verbs and Sentences (Thompson, 2011), - = not completed. [1] Normative data for PPA (Hsieh, Schubert, Hoon, Mioshi, & Hodges, 2013. Cut-off at overall score 82 indicates 100:1 likelihood of dementia Mioshi, Dawson, Mitchell, Arnold, & Hodges, 2006). [2]Normative data based on healthy sample aged 20-63 (Mansur, Carthery-Goulart, Bahia, Bak, & Nitrini, 2013). [3]Adapted from the NNB normative data for 37 individuals with PPA aged 48-81 (Thompson & Weintraub, 2014).

Individualised Cognitive Scaffolds

Each participant was assessed on cognitive measures pre-intervention (see Table 3), with performance used to inform cognitive scaffolds implemented in the intervention. The tests of verbal episodic memory and visual recognition memory, along with the ACE-III, were repeated post-intervention to monitor cognitive performance and where gains due to intervention or learning were not anticipated.

Executive function, specifically, inhibition, was measured on the Stroop Test (Stroop, 1935) to assess individual ability to suppress attention of competing stimulus during intervention. Visual attention was measured on the Trail Making Test (Tombaugh, 2004). The visual elevator subtest of the Test of Everyday Attention (Robertson et al., 1994) was used to assess individual ability to shift between tasks during intervention. All participants, with the exception of P1, demonstrated difficulty completing at least one component of the Stroop Test. Two participants diagnosed with svPPA (P2, P3) showed difficulty on all components, which may have been impacted by recall of colour names (i.e. impaired semantic knowledge). Participants diagnosed with AD showed impaired task shifting. P1 and P4 performed within normal limits, while the other participants diagnosed with svPPA (P2, P3) showed moderate deficits. All participants diagnosed with lvPPA demonstrated difficulty with task shifting, particularly P6 and P8. Three AD participants (P10, P11, P12) were unable to complete the attention shifting tasks. Scaffolds specific to executive function and visual attention were implemented in intervention for all participants, with the exception of P1 (svPPA) who demonstrated spared performance across all tasks.

Working memory was measured on the digit span and letter-number sequencing subtests of the Wechsler Memory Scale third edition (Wechsler, 1997) to assess individual ability to retain verbal instructions during intervention. The majority of participants with PPA performed within normal limits on the digit span task, with notably high scores for two svPPA participants (P1, P4). Three participants diagnosed with AD (P10, P11, P12), however, showed impaired performance. On the second working memory task, all participants showed difficulty, again, with the exception of P1 and P4. Scaffolds specific to working memory were implemented in intervention for all participants, with the exception of these two participants.

Verbal episodic memory was measured on the Hopkins Verbal Learning Test (Brandt, 1991) to assess individual ability to recall information and verbal stimuli during intervention. Impairments in verbal episodic memory were found for three participants diagnosed with AD (P9, P11, P12). Two participants diagnosed with svPPA (P2, P3) also showed difficulties, however, it is important to note that their performance was likely impacted by lexical deficits. Marginal deficits were seen for three participants with lvPPA (P6, P7, P8) and one with AD (P10). Scaffolds specific to verbal episodic memory were implemented in intervention for all participants diagnosed with AD, lvPPA (except for P5), and for two svPPA participants (P2, P3), although it is recognised that performance on these tasks may have been impacted by impaired semantic knowledge for participants diagnosed with svPPA.

Visual recognition memory was measured on the picture recognition subtest of the Rivermead Behavioural Memory Test (Wilson et al., 1985) to assess individual ability to process and retain visual stimuli during intervention. On the picture recognition task, P3 (svPPA) and two participants with AD (P11, P12) showed profound impairments, while P2 (svPPA) and two participants with lvPPA (P6, P7) showed moderate impairments in visual recognition. The remaining participants performed within normal limits, indicative of intact visual recognition. Scaffolds specific to visual recognition memory were implemented in intervention for two participants with svPPA (P2, P3), lvPPA (P6, P7), and AD (P11, P12)

Table 3

Pre-intervention assessment results of PPA and AD participants on cognitive testing to inform cognitive scaffolds

	Participant												Normative data
	1 svPPA	2 svPPA	3 svPPA	4 svPPA	5 lvPPA	6 lvPPA	7 lvPPA	8 lvPPA	9 AD	10 AD	11 AD	12 AD	*M (SD)*
Stroop													
Word score	100	29	40	85	60	81	83	96	51	44	56	7	103.8[1]
Colour score	61	25	21	59	42	47	56	36	27	30	17	4	80.6[1]
Colour-word score	40	-	-	36	22	6	23	5	15	13	5	-	48.4[1]
TMT – part A (seconds)	33	73	68	35	44	72	53	28	97	203	190	-	35.7(12.8)[2]
TMT – part B (seconds)	51	155	199	85	113	335	160	356	350	-	-	-	81.5(36.1)[2]
TEA – Visual elevator													
Accuracy (/10)	10	10	10	10	9	-	10	6	8	-	-	-	-
Timing (seconds)	3.4	6.3	5.6	2.8	5.6	-	10.1	15.8	12.7	-	-	-	-
WMS - III													
Letter-number sequencing (/21)	13	4	5	15	5	4	5	4	5	3	3	-	-
Digit span overall (/30)	18	12	15	24	10	13	13	12	18	11	6	8	-
HVLT													
Trial 1 (/12)	2	0	2	5	3	4	3	2	1	3	1	0	5.2(1.7)[3]
Trial 2 (/12)	6	2	1	6	8	4	4	5	3	4	2	2	7.1(2.1)[3]
Trial 3 (/12)	9	2	2	8	10	2	6	6	1	6	1	2	8.3(2.3)[3]
Delayed recall (/12)	6	0	0	6	7	5	3	3	0	7	0	0	6.9(3.2)[3]
Delay discrimination (/12)	10	2	0	8	10	9	9	7	9	7	0	0	9.1(2.2)[3]
RBMT													
Picture recognition (/10)	9	7	3	10	8	7	7	8	8	10	5	3	9.9(0.1)[4]

Note. TMT = Trail Making Test (Tombaugh, 2004), TEA = Test of Everyday Attention (Robertson, Ward, Ridgeway, & Nimmo-Smith, 1994), WMS-III = Wechsler Memory Scale third edition (Wechsler, 1997), HVLT = Hopkins Verbal Learning Test (Brandt, 1991), RBMT = Rivermead Behavioural Memory Test (Wilson, Cockburn, & Baddeley, 1985), - = not completed. [1]Normative data mathematically estimated from total time per hundred words to number of times in 45 seconds (Van der Elst, Van Boxtel, Van Breukelen, & Jolles, 2006). [2]Normative data based on healthy ageing sample (age *M*= 72.4, *SD*= 8.5). AD normative data; part A – 67.1(31.0), part B – 190.8(81.6) (Ashendorf et al., 2008). [3]Normative data based on Australian healthy ageing sample (age *M*=73.1, *SD*=5.6) (Hester, Kinsella, Ong, & Turner, 2004). [4]Normative data for control sample (age *M*=47.2), provided in RMBT manual.

Baseline Assessment and Outcome Measures

Three baseline assessments for naming performance and connected speech samples were conducted prior to intervention and then repeated again at one-week (initial post-intervention) and six-weeks (maintenance) after intervention to examine both treatment effectiveness and generalisation. Due to P4 living in a rural location, only one baseline assessment was completed for this participant.

Three baseline assessments of naming were repeated for picture stimuli which consisted of 144 items (48 nouns, 48 verbs, and 48 adjectives), sourced from non-copyrighted Internet material and guided by work on common conversational topics (e.g. Fried-Oken et al., 2015). Verbal responses were audio and manually recorded. Lexical items were balanced for frequency and length (number of syllables) across the three word classes and within treated and untreated items across all word classes for each participant. Data on the psycholinguistic variables of imageability and age of acquisition were also collected and monitored across treated and untreated sets for all participants (see Appendix C.1). As treatment sets were determined by accuracy for each individual at baseline testing, these were monitored on a case by case basis. Phonological errors or self-corrected responses were not penalised in scoring.

Three baseline assessments of connected speech were repeated which consisted of (a) nine everyday monologues from the Curtin University Discourse Protocol (Whitworth et al., 2015) including recounts (weekend, Christmas, past injury), procedures (changing a lightbulb, planning a meal, scrambling eggs), and expositions (global warming, obesity, bullying) (the nine topics were elicited over the three baseline testing points, with all three genres covered in each session); (b) a narrative monologue (Cinderella); and (c) two picture description tasks; Cookie Theft picture (Goodglass & Kaplan, 1983) and Birthday Cake picture (Nicholas & Brookshire, 1993). Samples were audio recorded using an iPad and were transcribed and coded for communicative informativeness (%CIUs) and efficiency (CIUs/min) (Nicholas & Brookshire, 1993) by the first author. Inter-rater reliability was established at >90%, differences were found across diagnostic groups and measures, with a trained speech-language pathologist on 50% of the samples.

Intervention Protocol

A strategic self-cueing approach was implemented for all participants over a six-week period. While this approach to lexical recovery drew on stimulation and relearning mechanisms, cognitive-relay was proposed to be the dominant mechanism of change (Beales et al., 2018), with the treatment including training of an internal cueing strategy for lexical retrieval (see Appendix C.2). In the initial two weeks, participants engaged in three sessions per week, reducing to two sessions per week in the following four weeks of intervention (total of 14 sessions). The duration of all therapy sessions was 60 minutes, with the exception of the 'communication partner prompted self-cueing' sessions, which were 90 minutes. Therapy sessions were conducted in the participant's home, with the exception of the rurally located participant (P4) who travelled to the city-based clinic rooms. Treated and untreated items were drawn for each individual from the 144 items in the baseline naming assessment (72 treated items and 72 untreated items) and matched for pre-intervention accuracy. Each participant and family member identified topics of high relevance from the established training stimuli. Stimuli were clustered within 12 semantic/thematic topics to capitalise on semantic relatedness to support recall, with one topic targeted each week for each participant.

Direct instruction and orientation to treatment was presented at the beginning of all sessions, which outlined the purpose and content of the intervention through verbal and visual modalities, covering (a) explicit teaching of the nature and processes involved in word retrieval, and (b) introduction and modeling of the components of the self-cueing strategy with reference to the strategic self-cue cards (see Appendix C.3). The protocol consisted of three phases:

Phase 1 "Clinician-prompted self-cueing" of intervention (six sessions) involved strategy prompting by the clinician and implementation of cognitive scaffolding to support participant engagement with the self-cueing strategy. The cognitive scaffolds (see Appendix C.4) were tailored to each participant following cognitive testing, with the exception of autobiographical memory scaffolds, which were utilised for all participants. The self-cue cards consisted of a range of questions covering semantic cues, e.g. 'what does it look like?', autobiographical memory cues, e.g. 'do you go to this place?', orthographic cue, e.g. 'can you write the first letter?', and phonological cue, e.g. 'what sound does the word begin with?'. Two

sessions each week focused on self-cueing of treated items in isolation, with the remaining session directed to self-cueing in the context of connected speech and promoting use of the strategy for any vocabulary (i.e. novel words that occurred naturally) when lexical retrieval difficulties occurred. Thematic topics (e.g. gardening) were prompted by the clinician to elicit a range of discourse genres. The clinician probed with the following question throughout all sessions "when you cannot think of a word what should you do?", Response: e.g. "Look at my cue card." Prior to commencing Phase 2 of intervention, participants were required to respond to this probe with a minimum of 80% accuracy, demonstrate evidence of independent attempts to self-cue, and demonstrate a retrieval accuracy of 70% or greater for treated items.

Phase 2 "Independent self-cueing" of intervention (four sessions) promoted independent self-cueing. The same structure and content was followed as Phase 1; however, this was integrated into one session per week, where half of the session focused on self-cueing in isolation and half on any vocabulary when lexical retrieval difficulties occurred in connected speech. For participants who did not respond to the self-cueing probe for a minimum of 80% accuracy, Phase 1 was continued and integrated into one session per week. Phase 2 differed from the earlier phase in that the clinician reduced strategy prompting and implementation of cognitive scaffolding.

Phase 3 "Communication partner prompted self-cueing" of intervention (four sessions) ran parallel with Phase 2 (or Phase 1 if continued) and required a consistent family member to attend. These sessions (one session per week) targeted use of the self-cueing strategy for the retrieval of novel vocabulary when lexical retrieval difficulties occurred in conversations, based on thematic topics. Treated and untreated stimuli were not purposefully targeted in connected speech. During conversations, the clinician modeled communication supports and prompts to educate the family member to facilitate the participant's use of the self-cueing strategy. Salient thematic topics were prompted, and personal items used to stimulate conversation (e.g. photographs, magazines, newspapers) when made available by the participant or family member. While the clinician engaged as necessary, this phase was primarily an opportunity for the family member to support and prompt the participant's strategy use.

To increase dose of strategy practice and facilitate use in everyday conversations, all participants engaged in home practice involving two activities. The first activity involved spoken naming practice of treated items using the picture stimuli and self-cue cards, with participants encouraged to complete this a minimum of 30 minutes, twice a week. The second activity required participants to use the self-cueing strategy while engaged in a conversation with their communication partner, reinforcing the transfer of strategies in interactions outside the intervention context, and further facilitating opportunity for strategy use with any vocabulary. Conversation practice was encouraged a minimum of 30 minutes each day of a week. Participants or communication partners were asked to record all occasions of practice, including documentation of the words practiced and conversation topics (see Appendix C.5).

Data Analysis

A series of Generalised Linear Mixed Models (GLMMs) were implemented through SPSS 24.0 (SPSS Corp., 2016). The GLMM represents a special class of regression model that accommodate dependent variables with markedly non-normal distributions, and that it includes both random and fixed effects. Time post diagnosis was not a confounding variable in any of the GLMMs and therefore was not included as a covariate in any of the models. One series of GLMMs were tested in order to determine the main and interactive effects of condition (treated and untreated), diagnosis (svPPA, lvPPA, AD), and time (baselines 1, 2, and 3, initial post-intervention, and maintenance) on naming accuracy for nouns, verbs, and adjectives. The GLMM for each of the three word types included one nominal random effect (participant), two nominal fixed effect (condition; diagnosis), one ordinal fixed effect (time), three 2-way interactions (Condition x Diagnosis, Diagnosis x Time, Condition x Time), and one 3-way interaction (Condition x Diagnosis x Time). A second series of GLMMs were tested in order to determine the main and interactive effects of diagnosis and time on connected speech performance in relation to everyday monologue, narrative, and picture description. The performance measures were %CIUs and CIUs/min. Each of the six GLMMs included one nominal random effect (participant), one nominal fixed effect (diagnosis), one ordinal fixed effect (time), and one 2-way interaction (Diagnosis x Time). The traditional ANOVA repeated measures model requires the following assumptions to be satisfied:

normality, homogeneity of variance, and sphericity. The GLMM 'robust statistics' option was invoked to accommodate violations of normality and homogeneity of variance. Violations of sphericity were accommodated by changing the covariance matrix from the default of compound symmetry to autoregressive. Compared to the traditional statistical procedures for analysing behavioural change, GLMM is less sensitive to participant attrition because it does not rely on participants providing data at every assessment point; the GLMM maximum likelihood procedure is a full information estimation procedure that uses all the data present at each assessment point. This reduces sampling bias and the need to replace missing data. GLMM is robust to unequal group sizes and is particularly powerful when group sizes are small. GLMM is able to use the data present at each assessment point because time is interpreted as a Level 1 variable that is nested within participant at Level 2.

Participants' data were also analysed individually through a series of pairwise comparisons between assessment periods (baseline and initial post-intervention, baseline and maintenance), word condition (treated and untreated), and word class conditions (noun, verb, and adjective), using the McNemar test to identify where significant differences occurred. To maintain a family-wise alpha rate of 0.05 over multiple comparisons, the Bonferroni adjustment was used for all pairwise comparisons. To determine changes in connected speech, the Fisher's Exact test was used to compare performance on communicative informativeness (%CIUs) and efficiency (CIUs/min) between assessment periods (baseline and initial post-intervention, baseline and maintenance), and discourse genre/task (everyday monologue, narrative, and picture description). A Poisson distribution was used in the communicative efficiency analyses. All *p* values for the McNemar and Fisher's Exact tests are reported in Appendix C.6, C.7, and C.8.

Results

All participants completed 14 sessions of therapy, with the exception of P7 (lvPPA) who completed eight sessions due to her availability. Ten of the 12 participants had a family member who participated in Phase 3; family members of two participants with PPA (P3, P7) elected not to participate. Eight participants progressed to Phase 2, with two svPPA participants (P2, P3) and two AD participants (P11, P12), not progressing through all stages. For these four participants, Phase 1 was extended and ran parallel to Phase 3. Eight participants reported completion of

home practice, with three participants not completing the conversation activity (P2, P3, P7), and P1 not completing either of the activities.

Aggregated Baseline Naming Data

Stability across the baseline period for all word classes was assessed and showed no significant change over the three baseline testing points, supporting the use of an aggregated baseline (see Appendix C.9).

Diagnostic Group Outcomes for Spoken Naming

For all word classes combined, there was a significant 3-way interaction between condition (treated, untreated), diagnosis (svPPA, lvPPA, AD), and time (baseline, initial post-intervention, maintenance) (F[4,54] = 15.062, $p < .001$, partial eta-squared = .53). Simple 2-way interactions between condition and time were subsequently analysed for the three diagnostic groups, with performance means for spoken naming for all, and individual, word classes presented in Table 4. There was a significant condition and time interaction for svPPA participants (F[2,18] = 5.706, $p = .012$, partial eta-squared = .31). For treated items, follow-up pairwise comparisons across the time factor showed significant improvements in naming performance from baseline to initial post-intervention (t[18] = 3.167, $p = .005$) and from baseline to maintenance (t[18] = 2.486, $p = .023$). For untreated items, there was a significant improvement in naming performance from baseline to initial post-intervention (t[18] = 2.536, $p = .021$) and from baseline to maintenance (t[18] = 6.596, $p = .035$). There was a significant condition and time interaction for lvPPA participants (F[2,18] = 46.961, $p < .001$, partial eta-squared = .84). For treated items, follow-up pairwise comparisons across the time factor showed significant improvements in naming performance from baseline to initial post-intervention (t[18] = 19.956, $p < .001$) and from baseline to maintenance (t[18] = 5.698, $p < .001$). For untreated items, there was a significant improvement in naming performance from baseline to initial post-intervention only (t[18] = 5.811, $p < .001$). There was a significant condition and time interaction for AD participants (F[2,18] = 11.352, $p = .001$, partial eta-squared = .56). For treated items, follow-up pairwise comparisons across the time factor showed significant improvements in naming performance from baseline to initial post-intervention (t[18] = 5.048, $p < .001$) and from baseline to maintenance (t[18] = 53.707, $p = .002$). For untreated items, there were also

significant improvement in naming performance from baseline to initial post-intervention (t[18] = 5.545, p < .001) and from baseline to maintenance (t[18] = 53.707, p = .002).

Table 4
Performance means for spoken naming by condition, diagnostic group, and assessment time

Condition	Diagnosis	Word class	*n*	Baseline	Initial post	Maintenance
Treated	svPPA	All words	72	19.832	32.750	30.000
		Nouns	24	7.750	12.250	11.000
		Verbs	24	9.499	13.500	12.500
		Adjectives	24	2.583	7.000	6.500
	lvPPA	All words	72	32.534	50.750	46.750
		Nouns	24	13.617	20.250	18.250
		Verbs	24	12.000	17.750	15.500
		Adjectives	24	6.917	12.750	13.000
	AD	All words	72	15.417	27.500	23.000
		Nouns	24	8.083	12.500	11.250
		Verbs	24	5.500	9.500	8.250
		Adjectives	24	1.833	5.500	3.500
Untreated	svPPA	All words	72	17.250	27.250	27.250
		Nouns	24	8.250	11.250	11.500
		Verbs	24	6.917	11.500	11.250
		Adjectives	24	2.083	4.500	4.500
	lvPPA	All words	72	32.164	45.250	39.000
		Nouns	24	13.583	17.500	16.750
		Verbs	24	12.414	16.750	15.250
		Adjectives	24	6.167	11.000	7.000
	AD	All words	72	13.717	21.500	22.000
		Nouns	24	7.667	10.000	11.000
		Verbs	24	4.217	7.250	7.250
		Adjectives	24	1.833	4.250	3.750

For the noun word class, a 3-way interaction between condition, diagnosis, and time interaction was not significant (F[4,54] = 2.432, p = .059, partial eta-squared = .15), and neither were the 2-way interactions between condition and time, diagnosis and time, or condition and diagnosis (all ps > .1). The main effect of time was significant; for both treated and untreated nouns, all three diagnostic groups showed significant improvements in naming performance from baseline to initial post-intervention (t[54] = 5.319, p < .001) and from baseline to maintenance (t[54] = 6.146, p = .002).

For the verb word class, a 3-way interaction between condition, diagnosis, and time was not significant (F[4,54] = 2.359, p = .065, partial eta-squared = .15). The condition and time interaction was significant (F[4,54] = 5.333, p = .008, partial

eta-squared = .28). For treated verbs, all three groups showed significant improvements in naming performance from baseline to initial post-intervention (t[54] = 10.444, p < .001) and from baseline to maintenance (t[18] = 4.896, p = .002). For untreated verbs, all three groups again showed significant improvements in naming performance from baseline to initial post-intervention (t[54] = 6.025, p < .001) and from baseline to maintenance (t[54] = 2.634, p = .011).

For the adjective word class, there was a significant 3-way interaction between condition, diagnosis, and time (F[4,54] = 5.466, p = .001, partial eta-squared = .29) for adjectives. There was no significant difference between treated and untreated adjectives over time for svPPA participants (F[2,18] = 1.333, p = .288, partial eta-squared = .13), although, combined, gains were significant (F[2,18] = 80.519, p < .001, partial eta-squared = .90). For combined treated and untreated adjectives, there were significant improvements in naming performance from baseline to initial post-intervention (t[18] = 10.690, p < .001) and from baseline to maintenance (t[18] = 3.732, p = .002). There was a significant interaction between condition and time for lvPPA participants (F[2,18] = 7.714, p = .004, partial eta-squared = .46). For treated adjectives, there were significant improvements in naming performance from baseline to initial post-intervention (t[18] = 4.509, p < .001) and from baseline to maintenance (t[18] = 3.218, p = .005). For untreated adjectives, there was a significant improvement in naming performance from baseline to initial post-intervention (t[18] = 5.607, p < .001) but not from baseline to maintenance (t[18] = 0.712, p = .485). Similar to participants with svPPA, the interaction between treated and untreated adjectives over time was not significant for AD participants (F[2,18] = 2.000, p = .164, partial eta-squared = .18). The effect of combined treated and untreated words was significant (F[2,18] = 5.732, p = .012, partial eta-squared = .39). For combined treated and untreated adjectives, there were significant improvements in naming performance from baseline to initial post-intervention (t[18] = 3.171, p = .005) and from baseline to maintenance (t[18] = 2.888, p = .005).

Individual Outcomes for Spoken Naming

For the noun word class, there was a significant improvement in naming treated nouns for two svPPA participants, specifically, P1 (McNemar test exact, p = .035) and P4 (p < .001) (see Table 5). These improvements were found to maintain

for both P1 and P4; $p = .035$ and $p = .019$, respectively. For P2, a significant improvement was found between baseline and maintenance ($p < .001$). No significant improvement for treated nouns was found for P3. For untreated nouns, a significant improvement was found for P1 between baseline and initial post-intervention ($p = .031$) and for P3 between baseline and maintenance ($p = .031$).

For participants with lvPPA there was a significant improvement in naming treated nouns for three of the four participants, specifically, P6, P7, and P8, between baseline and initial post-intervention, ($p < .001$ for all participants), which maintained for the three participants ($p = .011$, $p < .001$, and $p < .001$), respectively. No significant improvement for treated nouns was found for P5. For untreated nouns, a significant improvement was found between baseline and initial post-intervention for P6 ($p = .004$) and P8 ($p = .002$). Improvements between baseline and maintenance were found for P5, P7, and P8; $p = .004$, $p = .011$, and $p = .002$, respectively.

For participants with AD there was a significant improvement in naming treated nouns for three of the four participants, specifically, P9, P10, and P12; $p = .031$, $p = .002$, and $p = .008$, respectively, which were found to maintain; $p = .016$, $p = .002$, and $p = .004$, respectively. No significant improvement in naming treated nouns was found for P11. For untreated nouns, a significant improvement was found for P9 only between baseline and initial post-intervention ($p = .008$), which maintained ($p = .002$).

For the verb word class, there was a significant improvement in naming treated verbs for three svPPA participants, specifically, P1 ($p = .001$), P3 ($p = .016$), and P4 ($p = .031$). These improvements were found to maintain for each of the three participants; $p = .001$, $p = .016$, and $p = .035$, respectively. No improvements between assessment periods were found for P2. For untreated verbs, a significant improvement was found for P1 at initial post-intervention ($p = .016$) and maintenance ($p = .035$). Initial post-intervention effects were also found for P3 ($p = .031$), however, were not maintained. For participants with lvPPA there was a significant improvement in naming treated verbs for all participants, specifically, P5 ($p = .008$), P6 ($p = .001$), P7 ($p = .011$), and P8 ($p = .008$). These improvements were found to maintain for P5 ($p = .016$), P7 ($p = .001$), and P8 ($p = .035$). For untreated verbs, a significant improvement was found initial post-intervention for P6 ($p < .001$), P7 ($p = .016$), and P8 ($p = .001$), with maintenance of gains found for P8 only

(p = .019). For participants with AD there was a significant improvement in naming treated verbs for three participants, specifically, P9, P10, and P11; p = .019, p = .001, and p = .008, respectively, which were found to maintain for P10 (p = .006) and P11 (p = .031). No significant improvement for naming treated verbs was found for P12. For untreated verbs, a significant improvement was found for P9 and P10 at initial post-intervention; p = .019 and p = .016, respectively, which maintained for P10 only (p = .019).

Table 5

Individual participant significance values for naming performance on baseline to initial post-intervention and baseline to maintenance

			Nouns		Verbs		Adjectives	
			Treated	Untreated	Treated	Untreated	Treated	Untreated
SvPPA	P1	Initial Post	*	*	**	*	**	NS
		Maintenance	**	NS	**	*	*	NS
	P2	Initial Post	NS	NS	NS	NS	NS	NS
		Maintenance	***	NS	NS	NS	NS	NS
	P3	Initial Post	NS	NS	*	*	NS	NS
		Maintenance	NS	*	*	NS	NS	NS
	P4	Initial Post	***	NS	*	NS	**	NS
		Maintenance	*	NS	*	NS	**	NS
LvPPA	P5	Initial Post	NS	NS	**	NS	***	**
		Maintenance	NS	**	*	NS	***	*
	P6	Initial Post	***	**	**	***	*	*
		Maintenance	*	NS	NS	NS	NS	NS
	P7	Initial Post	***	NS	*	*	*	NS
		Maintenance	***	*	**	NS	NS	NS
	P8	Initial Post	***	**	**	**	**	NS
		Maintenance	***	**	*	*	**	NS
AD	P9	Initial Post	*	**	*	*	NS	NS
		Maintenance	*	**	NS	NS	NS	NS
	P10	Initial Post	**	NS	**	*	*	*
		Maintenance	**	NS	**	*	*	*
	P11	Initial Post	NS	NS	**	NS	NS	NS
		Maintenance	NS	NS	*	NS	NS	NS
	P12	Initial Post	**	NS	NS	NS	NS	NS
		Maintenance	**	NS	NS	NS	NS	NS

* p = < 0.05, ** p = < 0.01, *** p = < 0.001, NS = not significant

For the adjective word class, there was a significant improvement in naming treated adjectives for two svPPA participants, specifically, P1 (p = .008) and P4 (p = .002), which maintained for both participant; p = .031 and p = .002, respectively. No significant improvement for treated adjectives was found for P2 and P3. For untreated adjectives, no improvement at initial post-intervention or maintenance was

found for all participants. For participants with lvPPA there was a significant improvement naming treated adjectives for all participants, specifically, P5 ($p <$ .001), P6 ($p = .019$), P7 ($p = .031$), and P8 ($p = .008$). These improvements maintained for P5 ($p < .001$) and P8 ($p = .001$). For untreated adjectives, a significant improvement was found for P5 at initial post-intervention ($p = .001$) and maintenance ($p = .035$). A significant improvement was also found for P6 ($p = .019$), however, was not maintained. For participants with AD, one participant (P10) showed significant improvements in naming performance for treated verbs ($p = .016$) and untreated verbs ($p = .016$), with gains maintained for both conditions; $p = .016$ and $p = .031$. No significant improvements were found for P9, P11, or P12.

Diagnostic Group Outcomes for Connected Speech

Across level generalisation was examined through the main and interactive effects of diagnosis and time on connected speech performance in everyday monologue, narrative, and picture description. The diagnostic group means for communicative informativeness and efficiency are presented in Table 6. For everyday monologues, no significant 2-way interaction was found for either %CIUs ($F[3,24] = 1.673$, $p = .186$, partial eta-squared = .17) or for CIUs/min ($F[3,24] = 3.981$, $p = .848$, partial eta-squared = .33). Narrative analyses did not include three svPPA participants (P2, P3, P4), two lvPPA participants (P6, P8), and one AD participant (P12), as samples were unable to be elicited from these participants at all baselines, and therefore were not attempted post-intervention, due to difficulty comprehending the task instruction. For the remaining five participants, no significant 2-way interaction between diagnosis and time was found for %CIUs ($F[3,24] = 1.333$, $p = .255$, partial eta-squared = .14) or CIUs/min ($F[3,24] = 1.428$, $p = .227$, partial eta-squared = .15). For picture description, a significant 2-way interaction between diagnosis and time was found for %CIUs ($F[3,24] = 9.806$, $p = .040$, partial eta-squared = .55). A significant 2-way interaction was found for AD participants ($F[3,24] = 6.697$, $p = .001$, partial eta-squared = .46), while no significant interactions were found for svPPA ($F[3,24] = 2.750$, $p = .806$, partial eta-squared = .26) and lvPPA participants ($F[3,24] = 1.443$, $p = .247$, partial eta-squared = .15). Follow-up pairwise comparisons across the time factor showed a significant decrease in %CIUs between initial post-intervention and maintenance for AD

participants (p = .002). For CIUs/min, no significant 2-way interaction was found (F[3,24] = 1.367, p = .265, partial eta-squared = .15).

Table 6
Performance means for communicative informativeness (%CIUs) and communicative efficiency (CIUs/min) by discourse genre, diagnostic group, and assessment time

Measure	Diagnosis	Discourse genre	Baseline	Initial post	Maintenance
%CIUs	svPPA	Everyday monologue	59.000	57.750	65.000
		Narrative	11.468	9.968	12.968
		Picture description	54.250	58.000	60.500
	lvPPA	Everyday monologue	56.750	58.000	55.500
		Narrative	42.658	39.635	44.968
		Picture description	67.750	68.500	62.500
	AD	Everyday monologue	43.000	42.250	40.750
		Narrative	40.008	37.280	42.032
		Picture description	48.000	51.500	45.000
CIUs/min	svPPA	Everyday monologue	68.580	65.500	64.000
		Narrative	21.481	18.481	22.731
		Picture description	66.250	61.500	66.000
	lvPPA	Everyday monologue	57.250	54.750	56.250
		Narrative	48.631	54.131	50.131
		Picture description	58.250	65.750	49.750
	AD	Everyday monologue	45.000	47.500	48.000
		Narrative	34.269	29.519	32.769
		Picture description	40.750	45.000	40.250

Individual Outcomes for Connected Speech

There was a significant increase in %CIU in connected speech for two participants with svPPA (see Table 7). Specifically, P1 increased on the everyday monologue genre (from 58% to 73%) (Fisher exact, p = .019) and the picture description task (from 64% to 72%) (p = .011) at the initial post-intervention assessment. For P2, %CIU was found to increase between baseline and maintenance for everyday monologues (from 39% to 53%) (p = .033). Moreover, P2's %CIU was found to significantly increase on the picture description task at initial post-intervention (from 30% to 56%) (p = .001). No other increases in %CIU were found for svPPA participants. For participants with lvPPA, one participant (P5) showed increased %CIU on the narrative genre (from 76% to 87%) (p = .035), with improvements maintained (from 76% to 88%) (p = .022). No other significant changes in %CIU were found for participants with lvPPA. For participants with AD, one participant (P10) showed increased %CIU on the picture description task from

baseline (60%) to initial post-intervention (74%) (p = .026). No other significant changes in %CIU were found for AD participants.

Table 7

Individual participant significance values for %CIU and CIUs/min on baseline to initial post-intervention and baseline to maintenance

			Everyday Monologues		Narrative		Picture Description	
			%CIU	CIUs/min	%CIU	CIUs/min	%CIU	CIUs/min
SvPPA	P1	Initial Post	*	NS	NS	NS	*	NS
		Maintenance	NS	NS	NS	NS	NS	NS
	P2	Initial Post	NS	NS	-	-	*	NS
		Maintenance	*	NS	-	-	**	NS
	P3	Initial Post	NS	NS	-	-	NS	NS
		Maintenance	NS	NS	-	-	NS	NS
	P4	Initial Post	NS	NS	-	-	NS	NS
		Maintenance	NS	NS	-	-	NS	NS
LvPPA	P5	Initial Post	NS	NS	*	*	NS	NS
		Maintenance	NS	NS	*	*	NS	NS
	P6	Initial Post	NS	NS	-	-	NS	NS
		Maintenance	NS	NS	-	-	NS	NS
	P7	Initial Post	NS	NS	NS	NS	NS	NS
		Maintenance	NS	NS	NS	NS	NS	NS
	P8	Initial Post	NS	NS	-	-	NS	NS
		Maintenance	NS	NS	-	-	NS	NS
AD	P9	Initial Post	NS	NS	NS	NS	NS	NS
		Maintenance	NS	NS	NS	NS	NS	NS
	P10	Initial Post	NS	NS	NS	NS	*	*
		Maintenance	NS	NS	NS	NS	NS	NS
	P11	Initial Post	NS	NS	NS	NS	NS	NS
		Maintenance	NS	NS	NS	NS	NS	NS
	P12	Initial Post	n/a	n/a	-	-	n/a	n/a
		Maintenance	NS	NS	-	-	NS	NS

* p = < 0.05, ** p = < 0.01, *** p = < 0.001, NS = not significant, "n/a" data not available, "-" participant unable to complete assessment.

For participants with svPPA there were no significant changes in CIUs/min in connected speech following intervention. For participants with lvPPA, one participant (P5) showed increased CIUs/min on the narrative genre (from 57 to 82 CIUs/min) (p = .021), with improvements maintained (from 57 to 82 CIUs/min) (p = .021). For participants with AD, one participant (P10) showed increased CIUs/min on the picture description task from baseline (63 CIUs/min) to initial post-intervention (88 CIUs/min) (p = .025). No other significant changes in CIUs/min were found for svPPA, lvPPA, or AD participants.

Cognitive Control Measures

Analysis of cognitive control measures, using 2-way interactions, found no significant change in the ACE-III from pre-intervention to initial post-intervention for svPPA (F[1,15] = 0.154, p = .701, partial eta-squared = .01), lvPPA (F[1,15] = 1.733, p = .208, partial eta-squared = .10), or AD (F[1,15] = 0.097, p = .760, partial eta-squared = .01). No significant change in the test of verbal episodic memory was found for svPPA (F[1,15] = 2.361, p = .145, partial eta-squared = .14), lvPPA (F[1,15] = 1.029, p = .327, partial eta-squared = .06), or AD (F[1,15] = 0.444, p = .515, partial eta-squared = .03). Similarly, no significant change in the visual recognition memory test were found for svPPA (F[1,15] = .738, p = .494, partial eta-squared = .05), lvPPA (F[1,15] = 1.520, p = .573, partial eta-squared = .09), or AD (F[1,15] = 0.974, p = .860, partial eta-squared = .06) (see Appendix C.10).

Discussion

This study investigated the effectiveness and generalisation of a self-cueing lexical intervention for 12 participants with diagnoses of svPPA, lvPPA, and AD, examining performance of diagnostic groups on a range of measures that spanned naming and connected speech. Within level generalisation was actively facilitated by the combination of strategy use, individualised cognitive scaffolds, and increased dose and practice through involvement of the communication partner, while across level generalisation was targeted through incorporation of different word classes and practice of strategies with novel (untreated) but salient vocabulary in the connected speech environment of conversation with communication partners. Examination of individual performance highlighted variability, with promising findings of within level generalisation found, while no clear patterns emerged for across level generalisation.

Our findings showed direct treatment effects for all diagnostic groups, supportive of our hypotheses. A significant increase in retrieval of all treated word classes (nouns, verbs, and adjectives) was found, consistent with our prediction that the strategy approach would support lexical retrieval, irrespective of word class. Also consistent with our hypotheses, within level generalisation was found for all diagnostic groups and, similar to direct treatment effects, was found for all word classes. Significant improvements in naming treated and untreated items maintained at six-weeks post-intervention, with the exception of untreated adjectives for

participants with lvPPA. Although direct treatment effects and generalisation to untreated items was found for all word classes based on diagnostic group analysis, follow up analysis of individual data revealed that improvement of treated items was most commonly seen for verbs (10/12 participants), followed by nouns (9/12 participants), and adjectives (7/12 participants). Similar patterns for generalisation to untreated items were seen, with improvement most commonly found for nouns (10/12 participants) and verbs (7/12 participants), while untreated adjectives were found to improve for 3 participants. While across level generalisation to connected speech was expected for all diagnostic groups, no change was seen in communicative informativeness and efficiency in the discourse samples, involving picture descriptions and monologues across a range of genres, in any of the diagnostic groups. This will be explored further below. As predicted, no change in nonlinguistic cognitive measures was found for any of the diagnostic groups. This suggests that while the scaffolding of the cognitive skills may have served to support therapy, these processes did not respond in a therapeutic way through their implementation. The current study supports a growing body of evidence for direct treatment effects following lexical retrieval interventions for people with progressive lexical impairments, with significant improvements in words targeted in therapy seen across word classes and diagnostic groups. The presence of consistent and significant within level generalisation to untreated items across all diagnostic groups, contrasts with the previous PPA lexical retrieval intervention literature where treatment effects have been, more frequently than not, item-specific (Beales et al., 2018; Croot, 2018). The inclusion of participants with AD in a strategic lexical retrieval intervention with a multi-cue approach is also novel within the current AD literature.

Pre-Intervention Cognitive Profiles

Within each diagnostic group, there were participants who demonstrated consistent significant improvements across word classes, while others showed limited gains. Pre-intervention cognitive profiles may have had an impact on outcomes following intervention, including both within and across level effects. For participants with svPPA, strong evidence to support direct treatment effects were found for P1 and P4, while limited improvements were found for P2 and P3. Significant within level generalisation was seen for P1, with the exception of untreated adjectives in which gains did not generalise. Moreover, improvements in

%CIU were found at initial post-intervention on the picture description and everyday monologues task for P1. By contrast, P2 showed no improvements in naming with the exception of increased performance in noun naming at maintenance, while P3 showed improvements in verb naming only which generalised to untreated items. Despite limited gains in naming performance, P2 showed improvements in %CIU for picture description and everyday monologues, with maintained effects only for picture description. Interestingly, P1 and P4 showed within normal limits performance on the majority of pre-intervention cognitive measures including the domains of executive function and visual memory, working memory, verbal episodic memory, and visual recognition memory, in comparison to P2 and P3 who presented with impairments across all domains.

For participants with lvPPA, improvements in naming performance were seen across all word classes and treatment conditions with the exception of P7 and P8 who showed no generalised improvements for naming untreated adjectives and P5 who showed no gains in noun naming or generalisation to untreated verbs. It is important to note that on baseline noun naming, P5 scored close to ceiling which may have had implications for performance change. Interestingly, across level generalisation effects for %CIU and CIU/min were found for P5 only, with significant improvements found on the narrative task. In contrast to the svPPA diagnostic group, performance on pre-intervention cognitive measures showed similar outcomes between individuals with lvPPA. Specifically, all lvPPA participants presented with impaired executive function, visual attention, and working memory. On measures of visual recognition memory, P6 and P7 showed impaired performance, while P5 and P8 performed within normal limits. Importantly, P5 was the only lvPPA participant with intact verbal episodic memory. Given that P5 was the only lvPPA participant to show instances of across level change, a possible association between across level generalisation and verbal episodic memory capacity could be indicated. Moreover, P5 was the only lvPPA participant to show improved naming of untreated adjectives, note, P6 also showed initial post-intervention improvements, however, gains did not maintain.

Patterns between pre-intervention cognitive profiles and intervention outcomes are also seen for participants with AD. Improved performance in naming was most consistently found for P9 and P10, with instances of significant improvement found across all word classes and treatment conditions with the exception of adjectives (treated and untreated) for P9 and untreated nouns for P10. In

contrast, limited gains were found for P11 and P12, with improvements in naming performance seen for treated verbs only for P11 and treated nouns only for P12. P10, who showed the most consistent improvements in naming performance, was also the only participant with AD found to show instances of across level generalisation, with improvements seen in %CIU and CIU/min on the picture description task. Limited treatment and generalisation gains seen for P11 and P12 may have been associated with significant cognitive impairments across all domains. By contrast, P9 showed mild impairment across all nonlinguistic cognitive domains, which may have contributed to the gains seen for noun and verb naming. Interestingly, P10 was the only participant with AD to demonstrate within normal limits performance on verbal episodic memory and visual recognition memory. Similarly to P1 and P5, P10 showed instances of across level generalisation and demonstrated the most consistent naming improvements in comparison to other individuals within their given diagnostic group, highlighting the potential impact of nonlinguistic cognitive functions, particularly verbal episodic memory and/or visual recognition memory, on intervention outcomes found and consequently, capacity for strategy uptake.

Strategic Approach

The inclusion of a strategic approach, in contrast to, for example, repetition naming approaches, was suggested to have played a critical role in facilitating increased generalisation seen in our earlier study (Beales et al, 2016), and also potentially contributing to the across level generalisation that was found for three of the four participants in that study. In the post-stroke aphasia literature, it has been reported that generalisation to untreated items is increased when a strategy approach is applied (Nickels, 2002). In the current study, the inclusion of a strategy for lexical retrieval is also proposed to have contributed to the significant within level generalisation outcomes seen, however, it was not reflected in across level generalisation outcomes, a finding which was unexpected given the same discourse behaviours were used to measure this. Building on from the Beales et al. (2016) study, people with AD were also included here, and demonstrated treatment and within level generalisation outcomes similar to PPA variant groups. Given the trend of predominantly memory based therapies applied for lexical retrieval in AD, this study provides novel evidence to support the use of language based interventions, specifically, strategy approaches. Strategy practice, both in the current intervention protocol and home practice, encouraged use of the self-cueing strategy for any

vocabulary (i.e. novel words that occurred naturally in connected speech), aiming to enhance both salience of practice and transferability of the strategy. Evidence of significant improvements in untreated items were also evident across all word classes, reinforcing the importance of extending intervention focus from nouns and verbs to adjectives as well (Renvall et al., 2013).

Cognitive Scaffolding

In this study, we incorporated nonlinguistic cognitive scaffolding, based on pre-intervention assessment, to support participants' ability to engage in therapy as well as optimise their capacity to take on strategies for lexical retrieval. Due to the progressive nature of diagnostic groups in this study and the expectation that cognitive difficulties would evolve over time, we aimed to reduce the cognitive load during the intervention to ultimately optimise resources for lexical retrieval and facilitate uptake of strategy. Despite differences in the underlying nature of PPA and AD, we anticipated cognitive scaffolding to assist all diagnostic groups in the intervention. While not all cognitive processes were reassessed, those that were showed no significant change, suggesting that the scaffolding did not, as expected, act in a therapeutic way for the cognitive deficits. The contribution of the cognitive scaffolding, while difficult to quantify, may assist in explaining why within level generalisation was seen to the extent that it was. It is anticipated that the implementation of cognitive scaffolds enhanced uptake of the self-cueing strategy due to reduced cognitive load, supportive of greater capacity for therapy engagement. Future research with progressive lexical impairments might examine which cognitive functions, impaired and retained, may predict an individual's ability to learn strategies and thus, the degree of improvement following intervention, given that variability was seen at the individual level. Determining whether particular cognitive scores, particularly verbal episodic memory and visual recognition memory, are associated with language intervention outcomes and, if so, which cognitive functions might be more strongly predictive, may not only provide insight into candidacy for therapy (e.g. individual's strategic competence), but also inform whether participants would benefit from intervention designs that train specific nonlinguistic cognitive functions prior to language intervention.

Connected Speech Environment and Communication Partners

Through incorporation of self-cueing practice in the context of connected speech, we aimed to improve lexical retrieval in communicative activities that were

similar to real-life interactions. Given the extensive and interconnected nature of linguistic networks (Price, 2010), training lexical retrieval in connected speech contexts concurrently with lexical retrieval in isolation, may have attributed to the significant within level generalisation outcomes found in this study. In the current study, however, limited evidence to support across level generalisation to communicative informativeness and efficiency was found. It is important also to note that seven participants were unable to complete the narrative task, however, were able to complete the other genres.

Multiple factors may have impacted the lack of across level generalisation found in this study. In the context of generalisation, Whitworth et al. (2017), in the study with participants with PPA, had proposed that targeting lexical retrieval within discourse contexts, with an explicit focus on sentence and discourse structure, may be critical to facilitate greater lexical retrieval in everyday connected speech. Consequently, lack of across level generalisation in the current study may indicate that the incorporation of opportunities for strategy use in connected speech contexts is insufficient and that more systematic integration of words into sentences and discourse may be required. Macrolinguistic elements, as in the Whitworth et al. (2017) study, may need to be explicitly targeted, in addition to microlinguistic elements, in order for change to occur at the discourse level. This does not, however, explain why Beales et al. (2016) had found across level generalisation in the earlier study for svPPA participants. Across level generalisation outcomes may be related to the connected speech measures selected, specifically, communication informativeness and efficiency may have been reduced due to verbalised strategy use in connected speech. Moreover, if an individual's self-cueing is verbalised, rather than internalised, strategy use may impact these measures (e.g. increased circumlocution). Thus, alternative measures for strategy intervention approaches, such as word class counts, may be more representative of change at the connected speech level.

In addition to the use of connected speech contexts, the inclusion of communication partners is suggested as a key ingredient to the current intervention protocol for the enablement of increased opportunities for strategy use in the home environments, enhanced salience of topics and words practiced, as well as increased meaningful interactions. Consequently, evidence of within level generalisation is proposed to have been directly influenced by family members in facilitating strategy use for novel words in everyday conversations. The incorporation of personal stimuli

during Phase 3 may have also had implications, increasing the saliency of stimulated vocabulary, with the participant's home environment potentially also playing a role in these positive outcomes.

Limitations and Future Research

In building on the evidence related to generalisation and incorporating novel factors hypothesised to optimise outcomes, all within one intervention protocol, there was a risk of being unable to determine the relative contribution of factors of interest. To partially mitigate this, care was taken to implement an experimental design that controlled for as many factors as possible related to, for example, baseline performance, item sets, assessments used, structured intervention protocol, and monitoring of home engagement. In-depth examination of individual response to cognitive scaffolds, and opportunities offered at home by communication partners warrants further attention. Although this study reports an in-depth investigation, the small sample size is also a limitation, along with the heterogeneous nature of the diagnostic populations. The nonlinguistic cognitive profiles of participants with PPA and AD participants were varied, supporting the need for individual evaluation of cognitive functions to inform intervention design, as opposed to generic recommendations for specific dementia syndromes. In the current study, it is therefore difficult to draw conclusive recommendations as to the impact and/or contribution of cognitive scaffolding to improvements following intervention, a line of future research that would be enhanced through increased monitoring of cognitive performance and focused comparison of the inclusion and exclusion of cognitive scaffolding. Specifically, future research may directly train nonlinguistic cognitive functions prior to and/or alongside engagement in language intervention, as recommended in the post-stroke aphasia literature (e.g. Harnish & Lundine, 2015). In the traumatic brain injury literature, it has been reported that variability in participant characteristics makes it difficult to identify relevant candidacy factors for internal memory strategy interventions (O'Neil-Pirozzi et al., 2016). Future research may correlate performance on cognitive measures with therapy outcomes as predictors of therapy outcomes as well as in relation to disease severity. Finally, given the focus of this study, the issues related to discourse measurement also warrant mention. Different across level generalisation measures to those used in this study (i.e. communication informativeness and efficiency) may have better informed the outcomes of a strategy-based intervention. Connected speech measures, such as word

class counts, and monitoring of strategy use, may have more comprehensively explored across level generalisation effects.

Conclusion

This study examined the effects of a strategic self-cueing approach for people with PPA and AD and provided evidence of direct treatment effects (i.e. significantly improved picture naming of treated items) and within level generalisation (i.e. significantly improved picture naming of untreated items) for all diagnostic groups. Improved naming of treated and untreated items was found across all word classes, i.e. nouns, verbs, and adjectives, with varying patterns found on individual analysis which may be associated with pre-intervention cognitive profiles. While across level generalisation to connected speech was expected for all diagnostic groups, no change was seen in communicative informativeness and efficiency in the discourse samples, albeit some significant gains were seen on individual analysis. The heterogeneity of nonlinguistic cognitive profiles called for the implementation of cognitive scaffolding during intervention that was tailored to the individual. Family members were specifically involved to increase dose and saliency, and reinforce the approach in functional contexts. Further investigation is warranted to examine the effects of cognitive scaffolding, comparing this approach to active training of these functions. By considering deficits beyond linguistic abilities in intervention design, such research is likely to innovate more holistic speech-language pathology management of clients with progressive lexical impairments and continue exploring how to maximise the benefits of our interventions to functional communication.

Chapter 6

Chapter 6 presents the findings of the second study in Phase Three of the research that aimed to explore the perspectives and experiences of participants and their family members following participation in the strategic self-cueing intervention through thematic analysis of interview transcripts.

Study Overview

Although rapidly progressing, speech-language pathology intervention research in PPA and AD remains in its early infancy and is currently limited by lack of understanding of what people and their families feel about current services (Morello et al., 2017; Volkmer et al., 2020). In this study, the lived experience of participants and their family members, specifically in relation to their experience of intervention, is explored, pivotal to understanding the functional impact associated with the quantitative treatment outcomes reported in the previous study. As detailed in Chapter 5, a consistent family member was directly involved in the self-cueing intervention in order to target communication contexts relevant to the individual and enhance opportunities for strategy generalisation (Volkmer et al., 2019). Based on the positive impact of involving communication partners that has been well established in the post-stroke aphasia literature (e.g. Green, 1982), as well as knowledge that the majority of people with dementia receive care from their spouse at home (Kendig et al. 2010, Riedl et al. 2014), family member input was considered critical to both the intervention study as well as the qualitative evaluation reported in this study. Benefits of intervention of a qualitative nature were examined through thematic analysis of interview transcripts, which aimed to explore the perspectives and experiences of participants and their family members following participation in the intervention.

Beales, A., Bates, K., Cartwright, J., & Whitworth, A. (2019). Lost for words: Perspectives and experiences of people with primary progressive aphasia and Alzheimer's disease and their families of participation in a lexical retrieval intervention. *International Journal of Speech-Language Pathology*, *21*(5), 483-492.

International Journal of Speech-Language Pathology

ISSN: 1754-9507 (Print) 1754-9515 (Online) Journal homepage: https://www.tandfonline.com/loi/iasl20

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To cite this article: Ashleigh Beales, Kristyn Bates, Jade Cartwright & Anne Whitworth (2019) Lost for words: Perspectives and experiences of people with primary progressive aphasia and Alzheimer's disease and their families of participation in a lexical retrieval intervention, International Journal of Speech-Language Pathology, 21:5, 483-492, DOI: 10.1080/17549507.2019.1625439

To link to this article: https://doi.org/10.1080/17549507.2019.1625439

Published online: 17 Jun 2019.

International Journal of Speech-Language Pathology, 2019; 21: 483–492

Lost for words: Perspectives and experiences of people with primary progressive aphasia and Alzheimer's disease and their families of participation in a lexical retrieval intervention

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Abstract

Purpose: Previous qualitative research involving family members' experiences of living with a person with dementia has consistently revealed themes of reduced connectedness and reciprocity of communication, highlighting the importance of education, support and practical strategies to facilitate communication within families. This study aimed to evaluate the perspectives and experiences of both family members and people with dementia following participation in a targeted speech-language pathology intervention involving people with primary progressive aphasia (PPA) and Alzheimer's disease (AD) and their family members.
Method: Semi-structured interviews of eight people with dementia (six PPA, two AD) and 10 family members were conducted following an intervention to increase lexical retrieval within functional contexts. Thematic analysis was used to analyse the interview transcripts.
Result: Two themes common to participants with dementia and family members emerged: (1) perceived benefits of the intervention and (2) lack of previous information on communication difficulties. Two separate themes emerged for people with dementia, predominantly people with PPA, involving: (1) improved communication and (2) increased participation. Three separate themes emerged for family members: (1) increased awareness and knowledge, (2) increased value of interaction and engagement and (3) uncertainty of the future.
Conclusion: The findings of this qualitative study revealed a range of perspectives on the experiences of client and family participants following a communication focussed intervention, examining both the nature of perceived direct gains and gaining insight into the issues faced by these client populations and their families. The provision of individualised information and education should be a fundamental human right for all people with communication impairment with greater attention given to people with progressive conditions where such needs are not currently met.

Keywords: *primary progressive aphasia; Alzheimer's disease; qualitative research; family members; lexical retrieval intervention*

Introduction

Communication is consistently highlighted as an area of concern for people with dementia (PWD) and is a key contributor to caregiver burden (Pozzebon, Douglas, & Ames, 2016). Prominent language impairment is often seen in the dementia syndromes, primary progressive aphasia (PPA) and Alzheimer's disease (AD). Specifically, lexical retrieval difficulties evident in both PPA and AD, albeit with different aetiologies and anticipated trajectories of progression, can have an overwhelming impact on conversation and the success of interaction within families. Targeting these communication difficulties through accessible and systematic interventions that have validity to both the person with dementia and their family members is critical to overcome the challenges in communication and reduce their impact in daily life.

Using qualitative research methodologies, the perspectives and experiences of people with post-stroke aphasia (e.g. Brown, Worrall, Davidson, & Howe, 2012) and, to some extent, AD (e.g. Duggleby, Swindle, Peacock, & Ghosh, 2011), have been used to inform how health professionals, including speech-language pathologists, work with clients and families in clinical practice. Families of people with post-stroke aphasia have articulated their desire to be engaged in interventions that provide practical

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ISSN 1754-9507 print/ISSN 1754-9515 online
Published by Taylor & Francis
DOI: 10.1080/17549507.2019.1625439

education and training, to support meaningful relationships as well as independence and autonomy (Brown et al., 2012). With major communication challenges faced by families of PWD, the pursuit of novel interventions that both address and engage families in communication interventions is equally important, and yet exploration of client and family perspectives on the experiences, feasibility and satisfaction of such services (Riedl, Last, Danek, & Diehl-Schmid, 2014) has received little attention to date, particularly in relation to PPA.

Communication loss as a key experience of spousal caregivers

An understanding of client perceptions and experiences is critical to the development of tailored interventions as well as advocacy for service provision and funding (Prorok, Horgan, & Seitz, 2013). In a synthesis of 16 qualitative studies, the spousal caregiver experiences of living with a partner diagnosed with dementia captured, notably, a marked emphasis on themes related to communication (Pozzebon et al., 2016). "Loss of partner" emerged as the central theme, which reflected perceptions of reduced reciprocity and connectedness, loss of talking, and increased communication difficulties, ultimately impacting the quality of their dyadic interaction and relationship. This finding was consistent with reports within the dementia literature of communication as the greatest challenge experienced by spousal caregivers (e.g. Braun et al., 2009; Clare et al., 2012; de Vugt et al., 2003). Specific to PPA, in two studies involving single dyads, Pozzebon, Douglas, and Ames (2017), and Kindell, Sage, Wilkinson, and Keady (2014), explored the spouse and family member experience caring for a significant other with the semantic variant of PPA (svPPA). Thematic analysis of family interviews revealed a consistent theme of diminished and effortful conversation which, notably, lacked reciprocity. In Pozzebon et al.'s study (2017), the spouse reported feelings of inadequacy and guilt for not knowing how to support word-finding difficulties and failed communication attempts. Similarly, the spouse and son, in Kindell et al.'s (2014) study, reported that conversation, despite their best attempts, was minimal and rarely initiated by their family member with svPPA. In both studies, conversation was emphasised as one of the most devastating losses associated with dementia. With the majority of PWD cared for by family or friends in their homes (Kendig, Browning, Pedlow, Wells, & Thomas, 2010; Riedl et al., 2014), the importance of active engagement with significant others in the environment cannot be overstated. Understanding the perceptions and needs of the spouse, family member, and/or friend is not only essential to the success of targeted interventions but also to, ultimately, optimising and sustaining PWD to live at home.

Service delivery models for PWD and their family members

Group support programmes are the most frequently reported method of service delivery to meet the education and training needs of families to support communication difficulties associated with dementia, with both benefits and barriers evident in the literature. Banks, Rogalski, Medina, Skoglund, and Morhardt (2006) evaluated a series of education and support sessions for people with PPA and frontotemporal dementia (FTD) and their family members, which included presentations by social workers and clinical neuropsychologists, followed by support groups to allow discussion of the challenges of providing care for an individual with PPA or FTD. Participants reported positive feedback and this informed future presentations to involve collaboration with the speech-language pathology department for implementation of communication activities appropriate for individuals with PPA. People diagnosed with PPA and FTD were invited to attend if considered "high-functioning," however, this was left to the discretion of the respective family member. This may have limited what the study contributed in terms of the education and support needs of people living with PPA as it remains unknown as to whether people at later stages of disease progression may also have benefited and in what ways or needed different content. In a more recent study, Jokel et al. (2017) highlighted the need to consider the needs of both the people with PPA and the spouse/caregiver in designing education and support programmes. This intervention study consisted of five individuals with PPA and their spouses who participated in a 10-week programme. The programme comprised group counselling and education sessions for caregivers, language activities sessions for people with PPA, as well as combined group sessions in which communication strategies were practiced between spousal dyads. Specific recommendations are reported based on qualitative outcomes including the need to devote sessions to practicing communication strategies in spousal dyads, joint education sessions for people with PPA and their spouses from a range of professions, dedicated time for spouses to network with each other, as well as opportunities for individual language therapy (Jokel et al., 2017). Similarly, Morhardt, O'Hara, Zachrich, Wieneke, and Rogalski (2017) reported outcomes of a psycho-educational support programme for individuals with PPA and their partners. Informed by an initial pilot study, a formal intervention was conducted, comprising bi-monthly sessions of education, group discussion, and targeted activities. Initially, the programme included people with PPA only (six individuals in the pilot programme), however, this was extended to include their partner in the formal intervention (nine people with PPA and eight partners). Thematic analysis of observational field notes recorded for participants with

PPA by a research assistant was carried out; these were not recorded for partners. The pilot and formal intervention phases revealed similar themes including coping with limitations and language decline, dealing with increased dependency and experiencing stigma (Morhardt et al., 2017). In addition, resilience and a sense of belonging were identified as themes shared amongst participants, specifically, participants shared enthusiasm for sharing helpful compensation strategies for communication (e.g. communication notebooks). Although, they did not attend sessions, partner observations were collected at the end of the pilot programme. Partners agreed that a major benefit of the programme was the opportunity for their family member to meet others living with the same condition (Morhardt et al., 2017). Participants further expressed to their partners a preference for "hands on" sessions rather than fact-based presentations with readily applicable strategies. Input on communication by the speech-language pathologist was the most frequently mentioned session, with participants with PPA having reported that the strategies to help maintain and enhance communication provided them with a sense of hopefulness (Morhardt et al., 2017).

While these findings do highlight benefits of group support programmes, barriers to accessing such services have been reported. In a study of 86 people with younger onset dementia and their family members, Cations et al. (2017) reported that 66.7% of people chose not to use a formal community service despite this being recommended to 96.8% of participants. Analysis of interview data revealed multiple barriers to the access of services, such as dissatisfaction with programmes offered via aged care and difficulty relating to older members. Participants reported favourably on programmes that alleviated social isolation through meaningful engagement. These programmes were tailored to individual needs and that involved contribution to the community (Cations et al., 2017). In addition to care-receiver barriers, it has been reported in the literature that speech-language pathologists lack confidence in planning and delivering treatment to people with PPA (Taylor, Kingma, Croot, & Nickels, 2009). Exploration of client and family perspectives of their participation in support and intervention programmes may assist this.

While studies have highlighted participant preferences for individualised communication interventions which involve communication partners, few intervention studies delivered at an individual level have reported involvement of PWD and their families. Grasso, Shuster, and Henry (2017) reported on one of the few studies in the literature where caregivers in the home setting were trained to administer a lexical retrieval intervention following a period of intervention with a speech-language pathologist. Two individuals, one diagnosed with mild cognitive impairment and one with the logopenic variant PPA (lvPPA), and their partners, engaged in a treatment hierarchy that focussed on strategic retrieval of residual phonological, semantic and orthographic information. Significant improvements in lexical retrieval (i.e. picture naming accuracy) were found for both participants, with no significant differences found in the magnitude of treatment effects between the clinician- and caregiver-administered treatment phases. Following participation in intervention, a qualitative rating scale was administered to assess participant and caregiver perceptions regarding the effectiveness of treatment, in which positive changes were reported. Although, no interview data was collected, caregiver statements were reported which provided further insight into the perceived benefits from their participation. Specifically, improved ability to guide descriptions during naming difficulty and reduced frustration during communicative interactions when learned strategies were employed, were reported by caregivers (Grasso et al., 2017). These preliminary findings of positive benefits of participation in communication intervention warranted further investigation, with thematic analysis of interview data.

In summary, previous qualitative research of the spousal and family member experience of living with a significant other with dementia has consistently revealed themes of reduced reciprocity and connectedness, highlighting the need for speech-language pathology services that provide education, support and strategies to facilitate communication. An understanding of the perspectives and experiences of PWD and their families following participation in intervention is a necessary component here to both guide service delivery models and increase clinicians' confidence in treatment planning, with the ultimate aim of improving communication within the family, reducing the communicative burden on all involved, and maintaining PWD in their home environment.

Aims of the study

This qualitative study aimed to evaluate a home-based lexical retrieval intervention that involved PWD and their family members. The specific aims were to: (1) explore the perspectives and experiences of people with PPA and AD and their family members following an impairment-based lexical retrieval intervention; (2) explore any perceived benefits from participation in the intervention; and (3) inform future practice and service delivery models. All participants with PPA and AD had attended a six-week intervention programme, delivered by the first author three times weekly for two weeks and twice weekly for the remaining four weeks, aimed at training a self-cueing lexical retrieval strategy. A family member attended approximately one session per week where he/she received training on communication supports and prompts to encourage lexical retrieval strategies in conversation with the PWD. The speech-language pathologist (first author) modelled communication

supports and prompts to educate the family member to facilitate and enable retrieval during conversation with the person with dementia.

Method

Participants

Participants were recruited to the study via private speech-language pathologists, neurologists and geriatricians working in Perth, Western Australia, through use of flyers. Participants were recruited from rural, regional and metropolitan areas of Western Australia. A working diagnosis of PPA or AD was established in accordance with the current diagnostic criteria (Gorno-Tempini et al., 2011; McKhann et al., 2011) via the referring neurologist or geriatrician based on previous brain imaging and neuropsychological testing.

Demographic details of PWD

Eleven PWD were recruited to the intervention study. Interviews were attempted with all participants, however, due to comprehension deficits, three participants (one svPPA and two AD) were unable to complete the interview and their data not included. The eight participants who were able to complete the interview were aged between 60 and 86 years (mean age of 66.6 years). Two participants had a diagnosis of svPPA, four lvPPA and two AD see Table I for demographic information).

Demographic details of family members

Of the 11 PWD recruited to the wider study, 10 had a family member who participated in the intervention and post-intervention interview. For one PWD, the family member did not consent to participation in the intervention. Family members comprised nine spouses and one son (see Table I for participant relationship to family member).

Research protocol

Semi-structured interviews were conducted with participants and their family members one week following participation in an individualised lexical retrieval intervention. Interviews were conducted by an independent speech-language pathologist, blind to the participant's diagnosis and the intervention design, to obtain a detailed account of their perspectives and experiences of being involved in the intervention. Participants with dementia and family members were interviewed separately to allow opportunity for expression without interruption or influence. The interview questions (see Supporting Information Appendix A) were focussed around: (1) prior education and training regarding language difficulties associated with dementia; (2) perceptions and experiences of being involved in the intervention; and (3) perceived benefits and challenges of being involved in the intervention. While the interviewer was able to make short remarks or comments for clarification, these were kept to a minimum. All interviews were completed in the participant's home, with the exception of one participant and his spouse who lived in rural Western Australia and completed the interview in clinic rooms in Perth. Each interview lasted 10–20 min and was audio recorded and transcribed verbatim by the first author. Ethics approval for this study was received from the Curtin University Human Research Ethics Committee (HR218/2015).

Data analysis

A six-phase thematic analysis in accordance with Braun and Clarke's (2006) principles was used to analyse the interviews. Interview transcripts were read over three times by two authors (A.B., K.B) to enable initial identification of themes. Transcripts were checked against audio files to ensure accuracy and integrity of the interview data. Emerging ideas and themes from the interview data were coded independently across the entire data set and then discussed and agreed upon. Interview data and relevant quotes were collated for each transcript. Codes were pooled into initial themes and reviewed against coded

Table I. Demographic information on PPA and AD participants according to age, gender, diagnosis, education and family member relationship.

	PWD1	PWD2	PWD3	PWD4	PWD5	PWD6	PWD7	PWD8	PWD9*	PWD10*	PWD11*
Age (years)	62	72	60	70	59	86	60	64	55	68	58
Gender	M	M	F	M	F	M	F	F	M	M	M
Diagnosis	svPPA	svPPA	lvPPA	lvPPA	lvPPA	AD	AD	lvPPA	svPPA	AD	AD
Education (years)	18	14	16	14	9	9	13	10	16	12	16
Time post diagnosis (years)	2	1	3	3	1	1	2	1	3	4	4
Relationship to family member	Wife (FM1)	Wife (FM2)	Husband (FM3)	Wife (FM4)	Husband (FM5)	Wife (FM6)	Husband (FM7)	–	Son (FM8)	Wife (FM9)	Wife (FM10)

PWD: person with dementia; FM: family member; svPPA: semantic variant primary progressive aphasia; lvPPA: logopenic variant primary progressive aphasia; AD: Alzheimer's disease.
*PWD unable to participate in interview.
–Family member did not participate in intervention or post-intervention interview.

Table II. Type and frequency of themes common to people with dementia and family members.

Theme	People with dementia		
	PPA ($n=6$)	AD ($n=2$)	Family member ($n=10$)
Receptivity to intervention strategies	5	1	5
Improved communication support	2	1	4
Personalised strategies	2	1	2
Increased motivation and/or effort	3	1	5
Valued support	2	1	4
Lack of previous information on communication difficulties	4	2	10
Other services	4	2	4
Self-directed research	–	–	5
Personal contacts	–	–	1

PPA: primary progressive aphasia; AD: Alzheimer's disease.

Table III. Type and frequency of themes identified for people with dementia.

Theme	People with dementia	
	PPA ($n=6$)	AD ($n=2$)
Improved communication	5	1
Increased participation	3	–

PPA: primary progressive aphasia; AD: Alzheimer's disease.

Table IV. Type and frequency of themes identified for family member.

Theme	Family member (n = 10)
Increased awareness and knowledge	9
Increased value of interaction and engagement	7
Uncertainty of the future	5

extracts and the full data set to generate a thematic map. Theme names and definitions were reviewed in order to generate clear, accurate, and engaging themes. Agreement was reached for all theme names and quotes through discussion by all authors. Where differences occurred, the authors jointly reviewed quotes and discussed theme names and definitions until agreement was reached. Illustrative quotes from the interview transcripts are presented, along with the number of interview transcripts with which the themes arose.

Result

Themes that emerged that were common for both PWD and family members were identified and tallied, along with themes for the two groups independently. Two themes common to PWD and family members emerged from the interviews: (1) perceived benefits of the intervention, and (2) lack of previous information on communication difficulties, with sub themes present within each (see Table II). In addition, separate themes for both the experiences of PWD and family members emerged. Emergent themes for PWD comprised: (1) improved communication, and (2) increased participation (see Table III). For family members, the following themes emerged: (1) increased awareness and knowledge, (2) increased value of interaction and engagement and (3) uncertainty of the future (see Table IV).

Themes common to participants and family members

Perceived benefits of the intervention

Benefits from participating in the intervention were reported by six (75%) PWD and five (50%) family members, with the following subthemes: (1) improved communication support, (2) personalised strategies, (3) increased motivation and/or effort and (4) valued support. Specifically, three (37.5%) PWD reported improved support from their family members through use of strategies during conversation:

> *[Family member] gives me a word or expression that I can focus on it. In the past I just would not ask if I can't remember the word but now she helps me. (PWD1)*

> *Normally um [family member] would give me the answer or say the word for me straight up but um this has been really um careful and it's helped him and I love it. (PWD3)*

This enhanced communication support from family members was reciprocated, evident through four (40%) reports:

> *I've learnt how to help [PWD] with finding the words. It's helped [PWD] I think because there's a few incidence now I know where [PWD] been saying something and she couldn't get the word out and she knew how to prompt me to help find the word so I reckon that was very good. (FM5)*

The use of personalised communication strategies was also reported to be helpful for three (37.5%) PWD and two (20%) family members, allowing tailored support during conversations:

> *I think probably using the techniques of memory, personal memory, personal circumstances to either jog the conversation along or to help [PWD] to construct in that scenario so probably I've become more in tune with the use of memory and perhaps the first letter or the first syllable of a word. But probably the one that I've taken out of it most is personal memory. (FM9)*

Four (50%) PWD and five (50%) family members reported increased motivation and/or effort from their participation in the intervention. PWD reported that

they were practicing their communication strategies outside of therapy and were more motivated to improve:

> *Just doing it once doesn't come into your usual strategies and that sort of thing. So I need to practice um if I do that it will be right at my fingertips. And um I certainly do want to get the most out of it. (PWD3)*

One spouse reported that the intervention had given her partner with dementia motivation to practice communication strategies:

> *It's given him motivation to do something about it you know to find the words and all of that sort of stuff whereas before it was total frustration for him. (FM2)*

Additionally, family members reported that they themselves experienced increased motivation and effort during conversations with their significant other:

> *I need to do it more, I need to focus on it more so and more consciously creating interactions with us, consciously creating conversations with us, consciously reminding him of circumstances or memories to try and get him talking. It just comes back to the effort on our part and my part. (FM9)*

> *And probably making a more conscious effort to engage him in conversations because the biggest one of the biggest things when you're at home and you don't necessarily have to have a lot of conversation so it's um so it's making that effort. I know I need to make that effort. (FM10)*

Three (37.5%) PWD and four (40%) family members reported that they valued the support they received through participation in the intervention. Both PWD and family members reported feelings of hope and being understood:

> *I felt sort of like really relieved that somebody I can ask people cause she has given me a lot of information and she offers a lot of help. (PWD7)*

> *Just realising well it's a corny thing but that I wasn't on my own. (FM1)*

> *I've learnt that there is hope for such things and it's not all doom and gloom! (FM4)*

Lack of previous information on communication difficulties

Six (75%) PWD and all family members reported a lack of information about communication difficulties associated with dementia prior to their participation in the intervention, with the following sub themes: (1) other services, (2) self-directed research and (3) personal contacts. Six (75%) PWD reported that they had previously accessed services for information, however, was lacking information about communication or too general for their needs:

> *I know with my um neurologist he didn't really give me… I asked you know a few questions but um he didn't really… You know I said to him you know was there a reason that I have this and no. And then I said is there something I could do to improve my speech. Nothing. (PWD3)*

Four (40%) family members also shared feelings of dissatisfaction of prior information for communication difficulties received from services:

> *We did have some hints from the [staff at health facility] when they did the um neurological assessments on [PWD], at the completion of that they gave us a few hints and tips and some brochures and things like that and they were basic things like telling [PWD] to take your time, you know don't be embarrassed about stopping and continue on with the conversation. They were things like prepare yourself before you go in to something, keep a notebook, keep a diary. So there were some hints and tips that they gave us but they were basic. (FM7)*

Another spouse reported that his participation in the current study was the first time that he had understood the diagnosis and the associated communication difficulties:

> *We didn't really know what [PWD] had. [The therapist] was the first one to tell us exactly what she had. [The therapist] was the first person to explain the language problems. FM5*

Five (50%) family members reported self-directed research as their information source. Specifically, the Internet was a common source of information, often following advice provided by clinicians. One spouse (FM2) had received information from attending a dementia short course that she arranged herself in her capacity as a nurse. One spouse reported gaining information through personal contacts:

> *I did a bit of googling. There's information on it but I didn't really understand how it fit into dementia… like I didn't know the differences. I knew it was different to Alzheimer's but I didn't know the break down and where it sat in the big umbrella. (FM8)*

Themes identified by PWD only

Improved communication

Six (75%) PWD reported that their ability to find and remember words had improved, particularly through the application of communication strategies:

> *[Family member] notices that I'm getting more words now then I did before and how to go find them. I still have problems with them you know I get the first letter I get anything and she understands. (PWD2)*

Increased participation
Three (37.5%) PWD reported that they were joining in conversations and talking more following intervention:

> *So yeh I am joining in and some most sometimes I will go to someone and have a chat and you know people come to me comfortable to have a conversation. (PWD3)*

Themes identified by family members only

Increased awareness and knowledge
A prominent theme identified from family members was the awareness and knowledge that they had gained through participating in the intervention. Nine (90%) family members reported increased awareness of their significant other's communication strengths and weaknesses and improved understanding of how to support them:

> *We've talked about the absolute value of increased awareness. See I thought it wasn't there anymore, but what I've learnt is that some of it might still be and it may depend on how long or how much and usage and access, more or less that seems to be. So [the therapist] had terms like access and that was all helpful and fundamentally I came to see the sort of learning or relearning and I came to really understand the whole point of it. (FM1)*

> *If you live with someone with that sort of brain injury you kind of get desensitised to it and you forget that there's a deeper thing to this person and every now and then you get reminders. There's a lot more language there and understanding that my dad has that I wouldn't be exposed to without being a part of this. (FM8)*

Increased value of interaction and engagement
Seven (70%) family members reported that they valued the opportunity to participate in the intervention with their significant other. Family members reported that through participating they had the opportunity to notice communication strengths and weaknesses, try out strategies and interact with their significant other:

> *I've become more engaged with the whole issue and the timing was perfect. (FM1)*

> *It gave us all the opportunity to point out the things that um that we notice that we see with what's happening with [PWD] speech and gives us the opportunity to throw things in and try them and see how it goes and that's what it was all about I think. (FM7)*

> *For my part I'm quite sad that it's over because I think the exercise and the interaction I think has been beneficial in many different sort of ways. Perhaps not even just the ways that your recording but I think just the very act of the interaction and conversations. (FM9)*

> *I think I've learnt something about um trying to help [PWD] communicate better and having an opportunity to practice. (FM10)*

Uncertainty of the future
Five (50%) family members reported concerns going forward following the intervention. Family members reported concern regarding access to further information and services, particularly for assistance in planning for the future:

> *Dad's okay now health wise and independence wise but when he starts becoming less independent you know what do I need to put in place now. (FM8)*

> *I just don't know where to turn for anything else. (FM6)*

One family member reported that she would appreciate a consistent contact for information and follow-up sessions:

> *I would appreciate a 3 or 4 times a year thing with [the therapist] or somebody. I'm not looking for a support group yet but I have always been someone who wants to normalise or check out my impressions of anything to get some certainty and know that it's not just me, because I would want to deal with it if it were either the situation or me. (FM1)*

Discussion

Through the analysis of 18 interview transcripts of eight PWD and 10 family members, the perspectives and experiences of participants and family members were obtained following involvement in a home-based intervention that targeted lexical retrieval strategies to improve word-finding ability in everyday communication. Family members were involved in the intervention programme where lexical retrieval strategies were encouraged during conversation by the family member to facilitate lexical access when difficulties occurred. The findings of this qualitative study revealed a range of perspectives and insights into the experiences of all involved, providing a rich opportunity to understand engagement with the model of intervention and insights into issues faced by these client populations and their families.

Overall, both PWD and their family members responded positively to the intervention.

Perceived benefits of the intervention was a theme common to PWD and family members, in which benefits of improved communication support from family members, use of personalised communication strategies, and increased motivation and effort were identified. PWD and family members also reported that

they valued the support they received through participation in the intervention. In addition, improved communication and increased participation were key themes identified for PWD. The perceived benefits reported by PWD provide qualitative evidence to support the direct improvements following lexical retrieval intervention reported in the literature. Notably, benefits reported by people with AD were less than people with PPA. More benefits may have been reported by people with PPA in comparison to AD due to the language focus of the intervention, more applicable to focal language impairments seen in the early stages of PPA in comparison to AD where other cognitive processes such as memory are additionally affected (McKhann et al., 2011). The difference in diagnostic group sample size (i.e. 6 PPA and 2 AD) is likely also to have influenced the quantity and range of perceived benefits and further research is warranted. Despite growing evidence that shows improvements following lexical retrieval interventions for people with PPA (e.g. see Jokel, Graham, Rochon, & Leonard, 2014 for review), comments from family members highlighted the persistent difficulty for individuals with PPA to access impairment-based treatment for language difficulties as part of routine care provision. This is consistent with previous studies which have reported that people with PPA have difficulty accessing any speech-language pathology input (e.g. Taylor et al., 2009; Volkmer, Spector, Warren, & Beeke, 2018).

Perceived benefits, separate to PWD, were also reported by family members. Family members valued the opportunity to interact and engage with their significant other in the context of the intervention. The majority of family members (90%) also reported increased awareness and knowledge, particularly regarding their significant other's communication strengths and weaknesses, and how to support their communication difficulties. These perceived benefits endorse the inclusion of family members in intervention to capitalise on opportunities for interaction and education to enhance awareness. Family member involvement in intervention has implications for greater social connectedness, a consistent theme in qualitative research of the spousal caregiver experience of living with a partner with dementia (e.g. Pozzebon et al., 2016). Although, not directly explored in this study, the increased awareness and value of interactions may have implications for the meaningfulness and reciprocity of conversations, consistent themes reported in qualitative research of the spousal caregiver experience literature (e.g. Pozzebon et al., 2016). To determine whether perceived changes in communication practices are represented in interactions, analysis of dyadic conversations is required to build on these findings, comparing pre- and post-intervention dyadic conversations between PWD and their communication partner. Analysis of conversations, for example, for evidence of trouble-indicating behaviours (i.e. behaviours that flag that the PWD is experiencing difficulty in a conversation), repair behaviours, turning-taking and contribution measures (e.g. Taylor et al., 2014) to examine whether perceived changes, such as increased support from the family member and improved initiation from PWD, are represented in everyday conversation will provide important support for the findings of this study.

A prominent view and reported experience of both PWD and family members was centred on the lack of information they had received on the language difficulties associated with dementia prior to their participation in the intervention. While an opportunity was provided to participants to comment on this, its prominence in the data was high. Family member reports of self-directed research, particularly through use of the Internet, highlighted this unmet need. One family member reported that his participation in the intervention was his first contact with a speech-language pathologist and exposure to the communication implications of his partner's diagnosis. Given that all participants had seen a neurologist prior to recruitment to this study, it is known that participants and their families had received information about their diagnosis prior to their participation in the intervention programme. Consideration of the context and timing of information delivery is, however, warranted. As changes in language are the hallmark feature of PPA and often AD, access to a speech-language pathologist is critical for the PWD and family member/s to gain an understanding of the diagnosis, particularly important in the early stages of disease progression (McNeil & Duffy, 2001; Taylor et al., 2009), but also as the condition progresses. The increased awareness and knowledge reported by family members following intervention are endorsement of individualised treatment programmes as an effective context for education of communication changes associated with dementia syndromes, in addition to education and implementation of communication strategies. This is further evident in the reported value for the support received by PWD and family members through participation in the intervention. Further research is called for to explore the effectiveness of how and when information and education is provided to PWD and their family members. Whether information is most accessible when it is delivered in the context of assessment feedback sessions or group education conferences and presentations, or when provided during treatment to allow information to be tailored to the individual's areas of difficulty and needs, in a context that more closely reflects everyday interactions, are fundamental considerations when working with people with acquired neurological impairments. The findings do, however, reinforce the value of education that is both individualised and contextualised. This experience of information accessed and support received during the intervention underpinned an additional theme of

uncertainty of the future, specific to family members. Half of the family members experienced concerns going forward following the intervention, particularly in future planning and access to services and information. This was especially apparent for family members of people diagnosed with PPA, highlighting the reduced availability to patient management pathways in the community and a need for planned input that considers the needs of patient and families.

Limitations

As the methodology used in the study was a semi-structured interview format with interview questions structured around specific topics, the potential is always present to limit the focus of responses. The initial questions were, however, specifically structured to solicit information of types of information and training received, while the remaining questions focussed the interviewee on topics and provided opportunities to expand if these were relevant. While open-ended questions, for example, tell me about your experience of the intervention, may have elicited different themes, a decision was made to provide greater focus and to keep the topics consistent across the two groups of interviews. Further limitations considered for this study include sample bias as well as the results not being checked with the participants. By participating in the intervention, participants were tacitly receptive to the intervention prior to commencing, therefore, sample bias is a limitation. It is further recognised that, due to the relatively small sample size of the study and that participants varied across dementia types and PPA variants, generalisability of the findings to the wider AD and PPA population has limitations. As service availability and practices are also likely to vary across different states and countries, these findings, albeit from rural, regional and metropolitan areas of one Australian state, may not be generalisable to other contexts.

Conclusion

This study sought to explore the perspectives and experiences of an impairment-based lexical retrieval intervention for people with PPA and AD and their family members. This study reinforced the largely unmet need of providing PWD and their families with targeted information of the communicative consequences of their diagnosis. The provision of individualised information and education should be a fundamental human right for all people with communication impairment with greater attention given to people with progressive conditions where such needs are not currently met. Finally, the findings emphasise the importance of obtaining qualitative information about the perspectives, experiences and perceived benefits of participation in an impairment-based intervention that will likely influence future therapeutic endeavours and patient management pathways.

Acknowledgements

We would like to thank the participants and family members for their willingness to engage in the evaluation process.

Declaration of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the article.

Funding

This work was supported by the Dementia Australia Research Foundation and Curtin University through the Australian Government Research Training Programme.

Supplementary material

Supplemental data for this article can be accessed at http://dx.doi.org/10.1080/17549507.2019.1625439

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Chapter 7

Discussion

Chapter 7 summarises and integrates the key research findings in light of the research objectives and their links with, and contribution to, the literature on lexical retrieval interventions in PPA and AD. In this final chapter, a summary of the major theoretical and clinical implications is provided with suggestions for future research, and an outline of the key strengths and limitations of the research.

Summary and Integration of Major Research Findings

Lexical retrieval difficulties are an early symptom of PPA and AD (Gorno-Tempini et al., 2011; Taler & Philips, 2008), and can have profound implications for quality of life (Martyr et al., 2018; Pozzebon et al., 2016). Lexical retrieval impairment is a core diagnostic feature of both svPPA and lvPPA, present in both naming and spontaneous speech tasks (Gorno-Tempini et al., 2011). Lexical impairment in AD is often attributed to episodic or semantic memory loss (Bourgeois & Hickey, 2009). Additionally, impaired naming abilities have been attributed to degraded semantic memory and/or working memory (Braaten et al., 2006), resulting in impaired access to the semantic knowledge and/or phonological form (Salehi et al., 2017). Although rapidly progressing, speech-language pathology intervention research in PPA and AD remains in its infancy and is currently limited by our reduced understanding of the mechanisms through which intervention is effective and how we might facilitate generalisation of outcomes. Understanding the needs of people and their families and their access to services has also received little attention (Cadório et al., 2017; Manouilidou & Nerantzini, 2020; Morello et al., 2017; Volkmer et al., 2020). This program of research was driven by the desire to make a meaningful difference to the quality of life of people living with PPA and AD, whose everyday interactions are impacted by lexical retrieval difficulties. The main objective was to inform the development of proactive and rehabilitative-style interventions that maximise generalisation and aim for meaningful differences to communication and quality of life. To address the role of speech-language pathologists and inform best practice, exploration was needed to address the theoretical underpinnings, assessment, and implication of diagnostic features, along with the lived experiences of the people with PPA and AD and their family

members, specifically in relation to their experience of intervention. The research program comprised five lines of enquiry with independent aims, however, with interrelated outcomes and implications. In the following section, the major research findings of each study will be summarised and integrated, with later discussion of the implications and links to the current literature relevant to lexical retrieval interventions in PPA and AD.

In Phase One, the first study of this program of research was presented, which provided a foundation for the following studies through setting out the evidence base for existing PPA and AD lexical retrieval interventions. The specific aims of Phase One were to (1) identify which mechanisms of change have been applied to lexical retrieval intervention studies for people with PPA and AD, and whether particular mechanisms of change were associated with more effective outcomes, (2) determine whether particular mechanisms of change of lexical retrieval intervention were associated with within and across level linguistic generalisation, and (3) identify the role of nonlinguistic cognitive functions in the reviewed studies.

The findings revealed that there is no evidence yet to suggest that better treatment outcomes are associated more with one type of mechanism of change. Potentiality for change in lexical retrieval performance for people with PPA and AD is associated with both the recovery and compensatory change mechanisms, that is, stimulation, relearning, reorganisation, and cognitive-relay. While the relearning mechanism of change was the most prominent, it is not yet clear whether this mechanism is the most effective. Greater generalisation, within and across level, following strategy-based interventions (i.e. self-cueing treatments) was found, indicating a need for further consideration of cognitive-relay mechanisms. Exploration of metacognitive self-cueing designs is particularly critical in the AD literature, having only been considered in the PPA literature to date. A key conclusion from the review was that intervention designs would further benefit from the consideration of nonlinguistic cognitive functions, specifically, the manipulation of nonlinguistic cognitive functions to harness potential for uptake of strategies. Phase One concluded that no intervention studies to date had considered the potential implications of executive function, working memory, or attentional capacity, highlighting the need for further understanding of these nonlinguistic cognitive functions that may be affected in PPA and AD, and any subsequent impact on

intervention outcomes. Further, the lack of understanding of across level generalisation was identified as being limited by measurement of this level of change being highly variable across studies, limiting conclusions being drawn. Exploration of change beyond the treatment level (i.e. lexical retrieval in isolation) to language behaviours in functional contexts, such as the connected speech seen in everyday discourse, is critical to advance our understanding of the theoretical underpinnings of intervention and in making a meaningful difference to quality of life. The inherent difficulty of measuring connected speech that has been raised in the literature, particularly in relation to obtaining a representative sample of connected speech, was identified as requiring investigation.

In summary, the findings of Phase One, specifically, the limitations and gaps associated with determining best-practice speech-language pathology services for lexical retrieval in PPA and AD, informed the subsequent lines of enquiry in the research program. The need for understanding of the nonlinguistic cognitive functions in PPA and AD, as well as the investigation of the consistency of connected speech measures over successive sampling, are examined in Phase Two. Furthermore, the promising findings associated with self-cueing interventions informed the intervention design implemented and evaluated in Phase Three, as well as the implementation of cognitive scaffolding with the aim of manipulating nonlinguistic cognitive functions to enhance treatment outcomes.

Informed by the findings of Phase One, Phase Two of this research program addressed implications associated with the diagnostic criteria of PPA and AD, factors related to intervention design, and the assessment of change in connected speech following intervention. In the first study in Phase Two, the primary aims were to (1) examine sentence repetition in people with PPA and AD, using an error classification schema adapted from Hohlbaum et al. (2018), and (2) explore correlations with digit span abilities, tasks known to draw on verbal working memory. Findings from this study distinguished the svPPA diagnostic group based on sentence repetition and digit span performance, however, no clear classification patterns found for lvPPA and AD groups suggest limitations in the reliable use of this diagnostic criterion and the potential diagnostic overlap of these two dementia syndromes. Performance on verbal working memory tasks, evidenced by error patterns associated with all diagnostic groups reported, highlight implications for the design of language intervention. Specifically, working memory capacity will likely impact on an

individual's capacity for new learning and engagement in intervention and, accordingly, consideration is required regarding how treatment factors can be manipulated to minimise interference. Consequently, the interaction of nonlinguistic cognitive functions in language intervention requires consideration through direct training and/or scaffolding of identified impairments that may impact treatment outcomes, which are explored in Phase Three. Findings from Phase Two further revealed potential disruption in retrieval from secondary memory (i.e. storage of memory units which demand longer term storage from the primary memory) for people with svPPA. Explicit training of retrieval strategies in order to compensate for impaired semantic memory may support an individual's ability to search secondary memory on the basis of cues. This training may extend to retrieval strategies to support lexical retrieval through cognitive-relay mechanisms, such as strategic self-cueing, which were found to show promise for within and across level generalisation in Phase One of the research program.

The second study in Phase Two also addressed a critical assessment factor related to intervention as identified as a gap in the literature in Phase One, that is, the limited assessment of across level treatment outcomes. The specific aims of this study were to (1) examine the stability of connected speech over three consecutive weeks in people diagnosed with PPA and AD on measures of lexical content, fluency, and communicative informativeness and efficiency, and (2) examine stability of these measures in different discourse sampling tasks, specifically everyday monologues, narrative, and picture description, each with a view to furthering our understanding of the reliability of using discourse as an outcome measure. Relative stability of connected speech measures was reported, providing both a level of confidence in the sampling context, and preliminary evidence to support single baseline use, thereby warranting use as an intervention outcome in research and clinical practice. Sampling of connected speech at a single point during the baseline period may increase accessibility and promote measurement of across level generalisation in both research and clinical practice by reducing time demands. Improved accessibility to evaluation of generalised gains to connected speech in everyday discourse, through understanding of reliability of sampling as well as task and genre selection implications, provides preliminary evidence to support meaningful evaluation of change following intervention, and informed the use of across level generalisation measures in Phase Three.

Guided by the findings of the preceding phases, Phase Three evaluated the direct treatment and generalisation effects following a strategic self-cueing intervention, underpinned by the cognitive-relay change mechanism. The specific aims of this treatment study were to (1) identify direct treatment effects for spoken word naming for treated items, (2) explore generalisation outcomes for spoken word naming of untreated items (within level generalisation), and (3) examine generalisation outcomes in connected speech (across level generalisation) as measured by communicative informativeness and efficiency. The analysis of diagnostic groups indicated direct treatment effects, that is, significant improvements in the naming performance of treated items across all word classes for each of the two PPA groups and the AD group. Additionally, evidence to support the use of strategy-based lexical retrieval interventions for generalisation of improved naming to untreated items was found across word classes based on diagnostic group analysis. Follow up analysis of individual data indicated variation across diagnostic groups and word classes with respect to significant gains. Specifically, nine of the 12 participants improved on naming of treated nouns, 10 participants improved in naming treated verbs, and seven participants improved in naming treated adjectives. Similar to treated items, follow up analysis of individual data indicated variable outcomes for within level generalisation effects. Improved naming of untreated nouns and verbs was found for 7/12 participants, six of which (P1, P3, P6, P7, P8, and P9) were found to improve for both word classes, while P5 showed improvement for nouns only and P10 showed improvement for verbs only. For adjectives, within level generalisation to untreated items was seen for 3/12 participants (P5, P6, and P10). Despite predictions that strategy-use would generalise to connected speech, no change was seen in communicative informativeness and efficiency in the discourse samples, involving picture descriptions and monologues across a range of genres, at the diagnostic group level. In contrast, some evidence to support improvements in communicative informativeness and efficiency were found at the individual level, with improvements found for four participants. From the individual analysis, no clear patterns emerged for across level generalisation, that is, improvements were not found to be associated with select diagnostic groups, discourse genres, or measures. Specifically, P1 and P2 (svPPA) showed improvements in informativeness (%CIU) for everyday monologues and picture description, P5 (lvPPA) showed improvements

in informativeness and efficiency (CIUs/min) for narrative, and P10 (AD) showed improvements in informativeness and efficiency for picture description.

Evaluation of the strategic self-cueing intervention was extended to address the perspectives of participants and family members. This final study in Phase Three of the research program examined qualitative data from semi-structured interviews with specific aims to (1) explore the perspectives and experiences of people with PPA and AD and their family members following an impairment-based lexical retrieval intervention, (2) explore any perceived benefits from participation in the intervention, and (3) inform future practice and service delivery models. Overall, the self-cueing intervention was received positively by participants and their family members, with a range of perceived benefits identified. Of particular note, six (of the eight interviewed) participants reported improved communication following participation in the intervention. This suggests the need to consider alternative outcome measures with greater sensitivity to across level generalisation. Perceived benefits of intervention also included the use of personalised communication strategies and increased motivation and effort. Notably, benefits reported by people with AD were less than by people with PPA. More benefits may have been reported by people with PPA in comparison to AD due to the language focus of the intervention, and its applicability to focal language impairments seen in the early stages of PPA in comparison to AD where other cognitive processes such as memory are additionally affected. Moreover, the findings indicated a prominent view centered on the lack of information they had received on the language difficulties associated with dementia prior to their participation in the intervention, highlighting the need for the provision of individualised information and education related to communication impairment.

Theoretical and Clinical Implications and Future Research

In this section, the major theoretical and clinical implications, with links to the current literature relevant to assessment and treatment of lexical retrieval in PPA and AD, are discussed with the overarching aim to inform the broader context of managing language deficits in people with PPA and AD.

Individual and Diagnostic Variability. The variability reported between and within diagnostic groups, particularly based on analysis of individual data, evoke a range of theoretical and clinical implications. Overall, participants with lvPPA

demonstrated better capacity for within level generalisation in comparison to svPPA, consistent with the systematic review findings reported by Cadório et al. (2017). Limited within level generalisation in svPPA has been proposed to be associated with context-dependent learning, reliant on preserved episodic memory rather than semantic-based learning (Cadório et al., 2017). Preserved verbal episodic memory may account for the consistent gains for treated items seen for P1 and P4 across word classes, suggestive of item-specific learning through episodic memory rather than gains in the semantic system which would have a higher likelihood of generalisation effects. Reliance on episodic memory in svPPA does not, however, correspond to the generalised improvements seen for untreated verbs for P1 and P3, as well as nouns for P1. Implications of the self-cueing approach utilised in this research, as opposed to context-dependent learning, are described below. For individuals with lvPPA, promising patterns of within level generalisation were found despite impairments seen across executive function, visual attention, and working memory. Instances of with level generalisation were found for untreated nouns and verbs for all lvPPA participants, with the exception of verbs for P5, with gains in untreated adjectives also seen for P5 and P6. Again, these findings are supportive of Cadório et al. (2017), who proposed that better capacity for generalisation may be seen for lvPPA participants as impairments are not item-specific, and associated with preserved semantic networks. Additionally, within level generalisation for lvPPA participants may be attributed to the self-cueing approach and/or cognitive scaffolding, explored further below. For individuals with AD, similar to the PPA groups, variable outcomes of within level generalisation were found between participants. Specifically, P9 and P10 were the only participants with AD to demonstrate instances of within level generalisation, with improvements found for untreated nouns and verbs for P9 and improvements for untreated verbs and adjectives for P10.

Disease Severity and Cognitive Function. The variable performance found in the individual analyses raise question regarding the impact of disease severity on intervention outcomes. Although occasions of significant change were found for all participants, limited capacity for improvement was seen for two svPPA participants (P2 and P3) and two AD participants (P11 and P12). For these four participants, time post diagnosis was three years for P2 and P3, and four years for P11 and P12, in contrast to participants within the same diagnostic groups who ranged from 1-2 years

post diagnosis. Findings of reduced capacity for improvement associated with disease severity, a factor that may be reflected in time post diagnosis, have been documented within the literature Specifically, Robinson et al. (2009) found that reduced retention of learned skills was associated with disease severity, attributing to the notion that better scope for generalisation is seen for people who engage in intervention early in the disease course (Cadório et al., 2017). Disease severity and reduced responsiveness to therapy is likely further associated with the severity of cognitive functions. For the four participants described above (P2, P3, P11, and P12), impairment across all pre-intervention cognitive tests was found, with the exception of the visual attention subtest of the Test of Everyday Attention in which P2 and P3 (svPPA) performed within normal limits. Overall, P11 and P12 (AD) presented with increased severity of cognitive impairment compared to P2 and P3, with deficits inhibiting completion of a range of executive function, attention, and working memory tests. This contrast may indicate the contribution of factors additional to disease severity, which may be attributed to limited response to intervention for these two svPPA participants. Specifically, degraded semantic memory may have had additional implications to learning capacity, irrespective of strategic competence. Bier et al. (2009) proposed that new learning should draw on semantic memory, as opposed to episodic memory, in order to optimise generalisation within the semantic system. Interestingly, these four participants demonstrated the most impaired performance on verbal episodic memory comparative to the other participants in the study, which may have implications for strategy uptake. Moreover, time post diagnosis alone may not account fully for the limited gains following intervention seen, as P5 and P6 (lvPPA) were both three years post diagnosis at time of recruitment to the study, highlighting the need to consider factors of treatment responsiveness beyond disease severity.

Although further research is required to evaluate the impact of specific nonlinguistic cognitive functions (e.g. verbal episodic memory) on capacity for strategy uptake and response to intervention, the patterns drawn from the individual analyses suggest that spared cognitive functions may be vital to treatment outcomes. Parallels may be drawn with the post-stroke aphasia literature where greater improvement following intervention has been found for individuals with less impaired working memory and executive function (Fillingham et al., 2006), as well

as attention and visuospatial working memory (Harnish & Lundine, 2015; Villard & Kiran, 2015). Further investigation of the associations between an individual's nonlinguistic cognitive functions and strategic competence, would enhance interpretation of self-cueing strategy uptake and strengthen understanding of generalisation outcomes.

Episodic Memory and Strategic Competence. The individual analyses of treatment outcomes and pre-intervention nonlinguistic cognitive functions highlight the importance of episodic memory on an individual's strategic competence. The self-cueing approach involves strategy rather than repair, as participants are taught to prompt themselves without relying on others (Henry et al., 2013). According to Nickels (2002), this approach is suitable for participants with spared episodic memory. In the current research, implementation of a strategic self-cueing approach (i.e. cognitive-relay change mechanism), as opposed to context-dependent approaches that involve repeated naming in presence of a picture (e.g. stimulation change mechanism), may have capitalised on intact episodic memory and contributed to the promising findings of within level generalisation found. Preserved episodic memory, despite degraded semantic memory, may account for the generalised gains to untreated nouns and verbs seen for P1 (svPPA). Preserved episodic memory to optimise strategy uptake does not, however, account for the within level generalisation for verbs found for P3 (svPPA), who presented with episodic memory deficits. It is important to note that performance on the verbal episodic memory task for P3 may have been impacted by lexical deficits, warranting the need for additional measures of nonverbal episodic memory to inform the role of episodic memory in determining strategic competence. For individuals with lvPPA in this study, despite impairment seen across executive function, visual attention, and working memory tasks, only marginal deficits in verbal episodic memory were found for three lvPPA participants, with intact performance seen for P5. These findings highlight the potential role of episodic memory which may have attributed to the strategic competence of individuals with lvPPA in this study, as evidenced by findings of within level generalisation and progression to the independent self-cueing phase of intervention for all four participants.

Although impaired episodic memory is a typical feature in the early stages of AD, it is important to highlight that one AD participant (P10) showed marginal

deficits in verbal episodic memory, while significant impairments were seen for the remaining three. Interestingly, P10 demonstrated the most consistent within level generalisation in the AD diagnostic group, supporting the potential importance of episodic memory for uptake of strategies in intervention. The four participants, as discussed in the previous section, who demonstrated the most impaired performance on verbal episodic memory were also the only participants who did not progress to Phase 2 ("independent self-cueing") of intervention, providing further evidence to support the role of episodic memory in strategy uptake. Potential for within level generalisation found in this research is consistent with previous self-cueing interventions, albeit limited, reported in the PPA intervention literature where patterns of within level generalisation have been found for svPPA participants in addition to lvPPA participants (e.g. Beales et al., 2016; Henry et al., 2013), as reported in Phase One. The investigation of episodic memory function, inclusive of nonverbal tasks, with greater attention to control of these factors in study designing , is warranted in future research to determine the role that this cognitive function has on an individual's strategic competence, and importantly, treatment planning.

Cognitive Scaffolding. Analysis of individual data, as described above, revealed some cases of within level generalisation found for individuals with impaired episodic memory, as well as other cognition functions, raising questions regarding the role of cognitive scaffolds that were implemented during intervention. Despite recommendations in the post-stroke aphasia literature that nonlinguistic cognitive functions should be targeted in the context of language intervention, currently there has been limited investigation for progressive lexical impairments. In the intervention study, we incorporated nonlinguistic cognitive scaffolding, based on pre-intervention neuropsychological assessment, to support language processing during lexical retrieval. Due to the progressive nature of cognitive decline in each of the diagnostic groups in this study, we aimed to reduce the cognitive load impact during the intervention to ultimately optimise resources for lexical retrieval. It is important to note that, in addition to the four participants discussed above (P2, P3, P11, and P12), impairment of nonlinguistic cognitive functions were also found for P9 and P10 (AD), as well as all four lvPPA participants. For these six participants impairments across executive function, visual attention, and working memory domains were seen, albeit often marginal in nature. For these participants, it is

possible that the cognitive scaffolds utilised during intervention to compensate for impairments, tailored to the individual, may have influenced the improvements that were found. Importantly, the findings from this study highlight the critical need to evaluate cognitive functions pre-intervention and support the use of strategy training for individuals with intact cognitive functions.

In the current study, it is therefore difficult to draw conclusive recommendations as to the impact and/or contribution of cognitive scaffolding to improvements following intervention, a line of future research that would be enhanced through increased monitoring of cognitive performance and focused comparison of the inclusion and exclusion of cognitive scaffolding. Specifically, future research may directly train nonlinguistic cognitive functions prior to and/or alongside engagement in language intervention, as recommended in the post-stroke aphasia literature (e.g. Harnish & Lundine, 2015). The option of treating general cognitive abilities (e.g. working memory) is one of the recent advancements in language intervention, which has emerged from integrative accounts of language networks in the brain (e.g. Cahana-Amitay & Albert, 2015; Kiran & Thompson, 2019). Similar to post-stroke aphasia, individuals with neurodegenerative conditions are also predicted to have reduced capacity for cognitive resources which may attribute to language disruption. Consequently, training nonlinguistic cognitive functions, such as working memory, in PPA and AD may also result in positive transfer effects to linguistic domains, given that linguistic and nonlinguistic cognitive functions transpire from shared and distributed neural networks (Blumstein & Amso, 2013; Cahana-Amitay & Albert, 2014). The incorporation of cognitive targets highlights the need for investigation of multidisciplinary approaches, such as the interdisciplinary model proposed by Morhardt et al. (2015). Investigation of such intervention designs in neurodegenerative conditions, such as PPA and AD, would have further benefits in the theoretical advancements of understanding the language system as a broadly distributed neural network rather than specific domains of processing.

Active Ingredients in Intervention. In addition to the above participant characteristics and the impact on potentiality for change, the findings from this study reveal additional active ingredients of therapy that may have contributed to generalisation outcomes related to the therapy components and stimuli used. Active

ingredients are the elements of intervention which, when combined, are predicted to produce the intended intervention effects (Whyte et al., 2014). These factors are predicted to contribute to intervention targets, as opposed to inactive ingredients which are proposed to exert no effects on the intervention outcomes.

It has been suggested that the language component used in therapy should be dependent on the individual's underlying deficit, for example semantic consolidation in svPPA (Graham et al., 2001; Henry et al., 2008). Conversely, researchers have reported improvements in naming regardless of the component used. In the current strategic self-cueing intervention, multimodal training was targeted through incorporation of all cues (semantic, phonological, orthographic, and autobiographical information of the treated stimuli), with elaboration beyond simple repeated naming in an attempt to optimise treatment and generalisation outcomes (Reilly, 2015). In a systematic review of generalisation and maintenance effects following lexical retrieval interventions for PPA, Cadório et al. (2017) found promising long-term retention following intervention approaches that targeted deep semantic encoding, rather than repeated naming in presence of a picture (e.g. Savage et al., 2013). As reported in Phase Three, the majority of gains seen when assessed immediately post-intervention were maintained at 6-weeks follow up, inclusive of direct treatment and within level generalisation gains, which may be attributed to both the multimodal training and cognitive-relay change mechanism implemented in this research.

As reported in the literature review in Phase One, current PPA and AD lexical retrieval interventions have predominantly targeted noun stimuli. Similarities can be drawn with the post-stroke aphasia literature in which studies are typically limited to concrete nouns and verbs (Renvall et al., 2013). Building on a preliminary self-cueing intervention (Beales et al., 2016), nouns, verbs, and adjectives were implemented in this research to support access to a wider range of communicative intentions, and maximise facilitation of syntactic and argument structure.. Although direct treatment effects and generalisation to untreated items was found for all word classes based on diagnostic group analysis, follow up analysis of individual data revealed that improvement of treated items was most commonly seen for verbs (10/12 participants), followed by nouns (9/12 participants), and adjectives (7/12 participants). Similar patterns for generalisation to untreated items were seen, with improvement most commonly found for nouns (10/12 participants) and verbs (7/12 participants), while untreated adjectives were found to improve for 3 participants.

Evidence of significant improvements in treated and untreated items across all word types reinforce the importance of extending intervention focus from nouns, to verbs and adjectives as well (Renvall et al., 2013), however, caution should be taken with adjectives given the reduced outcomes for untreated items reported. Within level generalisation for adjectives was found for two participants with lvPPA (P5 and P6) and one participant with AD (P10), with no improvements found for svPPA participants. Lack of improvement of untreated adjectives for svPPA participants is inconsistent with the findings reported by Beales et al. (2016) who found generalisation to untreated adjectives for three svPPA participants. Beales et al. (2016) attributed this finding to transfer of the self-cueing strategy as well as the fluidity of adjectives and the noun links in the adjective semantic fields (Milman et al., 2014). Given the inconsistency between Beales et al. (2016) and this current research program, consideration of participant characteristics, inclusive of disease severity, and strategic competence, as addressed above, are needed. Further exploration of psycholinguistic variables and word class properties, such as imageability of treatment stimuli, as well as an increased sample size are warranted to better understand the comparatively reduced outcomes seen for adjective stimuli in this research and enable greater exploration of variability and identification of patterns.

In addition to word class types, Cadório et al. (2017) reported promising intervention outcomes for designs that adopted meaningful and familiar word stimuli, associated with the salience principle of neuroplasticity (Kleim & Jones, 2008). Several authors have reported the importance of relevant items, particularly for svPPA individuals due to optimisation of autobiographical memory as well as contextual links to enhance new learning and retention (e.g. Jokel et al., 2006; Robinson et al., 2009; Snowden & Neary, 2002). With regards to cognitive components, a small number of studies have used autobiographical information, as reported in Phase One of this research program, aiming to capitalise on spared memory systems (Jokel, Rochon, & Leonard, 2006; Snowden & Neary, 2002). In the current self-cueing intervention presented in Phase Three, direct treatment and generalisation outcomes may have been enhanced through incorporation of autiobiographical cues in the self-cueing approach in addition to inclusion of treatment stimuli and topics, guided by work on common conversational topics (e.g. Fried-Oken et al., 2015). Further exploration of word class effects as well as use of

functional and personalised stimuli, is warranted in future research to determine the role that these active ingredients play in determining generalisation outcomes as well as direct treatment effects.

Given the extensive and interconnected nature of linguistic networks (Price, 2010), training lexical retrieval in connected speech contexts and evaluation of across level generalisation was targeted in this research program, as presented in Phase Three, guided by the evidence of sampling stability and driven by the gap in measurement of across level generalisation reported in Phase Two. In the post-stroke aphasia literature, there has been some evidence to support training multiple processing levels to facilitate generalisation of treatment effects (e.g. Milman et al., 2014). Although there was some evidence to support improvements in communicative informativeness and efficiency found at the individual level, no clear patterns emerged for across level generalisation, that is, improvements were not found to be associated with select diagnostic groups, discourse genres, or measures. Consequently, the need for understanding discursive generalisation patterns following lexical retrieval interventions remains a critical gap for future research in progressive lexical impairments. From this research program, the need for a greater understanding of the connected speech genres and tasks used in clinical practice, as well as the use of linguistic measures themselves, is evident. This understanding will guide task and discourse genre selection, with specific consideration given to both the connected speech measures used and the purpose of language sampling (e.g. diagnostic profiling versus establishing baseline performance). In addition to the need for further consideration of the impact of discourse genre, research is also required to explore the stability and suitability of language measures in individuals with PPA and AD. Exploring the stability of other language measures (e.g. grammatical structures) may enhance current understanding about the language stability, and variability, in adults with PPA and AD. Lastly, additional measures to examine across level generalisation to those used in this study (i.e. communicative informativeness and efficiency) require exploration to inform outcomes of strategy-based interventions. Connected speech measures beyond communicative informativeness and efficiency, such as word class counts and monitoring of strategy use, require investigation to comprehensively explore the potentiality for across level change following strategic self-cueing approaches.

Key Strengths and Limitations

This research program, through the five interrelated studies, offers a novel and comprehensive contribution to the understanding and management of lexical impairments in PPA and AD. An in-depth investigation of individual participants and their respective diagnostic groups, combined with an examination of the aims of lexical interventions in progressive conditions, the implementation and evaluation of a novel treatment design aimed at optimising generalisation, and exploration of client/family perspectives of the intervention process, have provided an opportunity to deepen understanding of the potential for recovery in PPA and AD. While a novel, targeted lexical intervention was shown to be effective for all diagnostic categories represented here and for many of the individuals, the exploration of the underlying mechanisms of change and the therapeutic ingredients that may best facilitate both improvement and generalisation are regarded as the key contributions offered by this work and which inform a future research agenda.

Of particular note, while a framework of recovery and compensation was adapted for the mapping of the mechanisms of change within the PPA and AD lexical retrieval intervention literature, it is acknowledged that mechanisms of change may co-occur and that further investigation is warranted to explore how these may best be combined to maximise gains. The potential for combined and interacting mechanisms of change further support the need for explicit treatment designs with clear hypotheses regarding the underlying cause of change associated with the therapy approach. The enquiry into the dynamic interaction between cognitive and linguistic measures was a further prominent theme throughout the research program. Given the potential for cognitive scaffolding and capacity for improvement despite cognitive impairment found in this research program, the findings call for ongoing and systematic investigation of cognitive and linguistic mechanisms that impact both assessment and intervention of lexical retrieval, as well as other language difficulties, seen in PPA and AD. Of particular interest was the ability of some individuals to capitalise, to a greater degree, on strategic competence, suggesting that where cognitive processes such as episodic memory are intact, and/or cognitive scaffolding can be implemented to support marginal deficits, these processes may be used to support the learning of pro-active communication strategies such as self-cueing.

A number of limitations, however, have been acknowledged in the respective studies. The small sample size, along with the heterogeneous nature of the diagnostic populations, is a key limitation. Although GLMMs were employed to minimise these effects as far as possible given their robust handling of small data sets, the small sample size of the lvPPA, svPPA, and AD groups did impact the statistical power of group level analysis conducted across studies reported in this program of research. It is further recognised that, due to the relatively small sample size of the study and that participants varied across dementia types and PPA variants, generalisability of the findings to the wider AD and PPA population is reduced. While the rare nature of PPA does restrict recruitment, replication is recommended with a larger sample size. Further, as service availability and practices are also likely to vary across different states and countries, these findings, albeit from rural, regional and metropolitan areas of one Australian state, may not be generalisable to other contexts. It is also noteworthy that diagnoses in this study were not accompanied by postmortem confirmation or variant diagnosis for cases with AD. This may have particular implications for P10, who, although recruited to the research program with a diagnosis of AD, presented with some atypical features of AD (e.g. relatively preserved verbal episodic memory). P10's profile highlights the heterogeneity of cases, contributing to the emerging research which reports several unclassifiable cases, particularly related to the lvPPA and nfvPPA classification criteria (e.g. Sajjadi et al., 2012). Although P10 was diagnosed with AD by a neurologist specialised in the area of neurodegenerative diseases, this profile of AD may reveal an atypical presentation with implications for the linguistic AD variant of lvPPA (Vandenberghe, 2016).

Conclusion

This program of research aimed to provide insights into diagnosis, assessment, and intervention by seeking to deepen understanding of lexical retrieval impairment in people with PPA and AD. Through detailed profiling of cognitive and language behaviours, supported by neuroimaging data, and the design, implementation, and evaluation of a novel intervention protocol, this research program has informed implications for assessment and intervention in these clinical populations. The importance, for clinicians and researchers, of systematically profiling nonlinguistic cognitive functions in progressive conditions, with a view to

both deepening understanding of the conditions and informing subsequent intervention, is highlighted. Evidence to support rehabilitative-style interventions in PPA and AD, particularly in the early stages of the disease, is reported. These findings further highlight the potential impacts of disease progression and cognitive profiles, reinforcing the importance of early detection of the neuropathological changes in dementia (Synder et al., 2014), to support early intervention to optimise protective mechanisms against decline of everyday function in the early stages of disease. The perspectives of the participants and their families is equally critical to inform how interventions are delivered and how the benefits might be best measured, along with highlighting issues related to access and education. By seeking to combine knowledge of the critical ingredients needed to design and deliver language intervention with proposed recovery mechanisms and residual cognitive abilities, and in collaboration with the participants and their families, research is likely to innovate more effective and holistic speech-language pathology management of people with progressive lexical impairments and facilitate the generalisation of interventions to meaningful communication.

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(2) Manuscript iasl20:Determining stability in connected speech in primary progressive aphasia and Alzheimer's disease

Publication name and edition:

(1) International Journal of Speech-Language Pathology, Volume 21, 2019 - Issue 5

(2) International Journal of Speech-Language Pathology, Volume 20, 2018 - Issue 3

URL if applicable:

(1) https://doi.org/10.1080/17549507.2019.1625439

(2) https://doi.org/10.1080/17549507.2018.1442498

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Appendix B Human Research Ethics Committee approval

Curtin University

Office of Research and Development

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09-Jan-2018

Name: Anne Whitworth
Department/School: School of Psychology and Speech Pathology
Email: Anne.Whitworth@curtin.edu.au

Dear Anne Whitworth

RE: Annual report acknowledgment
Approval number: HR218/2015

Thank you for submitting an annual report to the Human Research Ethics Office for the project **Making the right connections: Assessment and treatment of language deficits in people with Primary Progressive Aphasia and Alzheimer-s disease**.

The Human Research Ethics Office acknowledges the project is ongoing and approval will remain current until 30-Nov-2018.

Any special conditions noted in the original approval letter still apply.

Standard conditions of approval

1. Research must be conducted according to the approved proposal
2. Report in a timely manner anything that might warrant review of ethical approval of the project including:
   - proposed changes to the approved proposal or conduct of the study
   - unanticipated problems that might affect continued ethical acceptability of the project
   - major deviations from the HREC approved protocol procedures and/or regulatory guidelines
   - serious adverse events
3. Amendments to the proposal must be approved by the Human Research Ethics Office before they are implemented (except where an amendment is undertaken to eliminate an immediate risk to participants)
4. An annual progress report must be submitted to the Human Research Ethics Office on or before the anniversary of approval and a completion report submitted on completion of the project
5. Personnel working on this project must be adequately qualified by education, training and experience for their role, or supervised
6. Personnel must disclose any actual or potential conflicts of interest, including any financial or other interest or affiliation, that bears on this project
7. Changes to personnel working on this project must be reported to the Human Research Ethics Office
8. Data and primary materials must be retained and stored in accordance with the Western Australian University Sector Disposal Authority (WAUSDA) and the Curtin University Research Data and Primary Materials policy
9. Where practicable, results of the research should be made available to the research participants in a timely and clear manner
10. Unless prohibited by contractual obligations, results of the research should be disseminated in a manner that will allow public scrutiny; the Human Research Ethics Office must be informed of any constraints on publication
11. Ethics approval is dependent upon ongoing compliance of the research with the Australian Code for the Responsible Conduct of Research, the National Statement on Ethical Conduct in Human Research, applicable legal requirements, and with Curtin University policies, procedures

Appendix C Chapter 5 Appendices

C.1 Naming stimuli synonyms and psycholinguistic variables

	Noun	Synonyms	Frequency*	Imageability*	Age of acquisition*	Syllables*
1	Flowers	-	1161	618	-	2
2	Plants	-	1408	605	-	1
3	Herbs	-	254	502	-	1
4	Lawnmower	-	24	-	-	3
5	Recipe	-	388	-	-	3
6	Barbecue	-	456	-	-	3
7	Blender	-	85	-	-	2
8	Microwave	-	199	-	-	3
9	Groceries	-	301	-	-	2
10	Butcher	Deli, Delicatessen	434	596	-	2
11	Bakery	-	179	-	-	1
12	Pharmacy	Chemist	175	-	-	3
13	Passport	-	534	546	-	2
14	Plane	-	4872	556	-	1
15	Museum	-	942	-	-	3
16	Tourist	-	237	577	-	2
17	Computer	-	3011	-	-	3
18	Internet	-	334	-	-	3
19	Camera	-	3011	576	-	2
20	Charger	-	62	-	-	2
21	Library	-	1170	587	-	2
22	Playground	-	260	-	-	2
23	Festival	Fete, Fair	505	-	-	3
24	Restaurant	-	2373	611	-	3
25	Birthday	-	4958	-	-	2
26	Camping	-	315	-	-	2
27	Pets	-	290	589	219	1
28	Grandchildren	Grandparents	198	-	-	3
29	Kitchen	-	2974	559	-	2
30	Laundry	-	972	-	367	2
31	Calendar	-	363	-	-	3
32	List	-	4110	-	-	1
33	Cricket bat	-	144	586	-	3
34	Stadium	-	312	586	-	3
35	Team	-	7528	565	-	1
36	Supporters	Fans	53	-	-	3
37	Storm	-	1574	587	-	1
38	Rainbow	-	407	604	-	2
39	Lake	River	1836	616	-	1
40	Flood	-	291	598	-	1
41	Racquets	-	3	522	-	2
42	Gym	-	927	613	-	1
43	Bike	-	1320	-	-	1
44	Sunscreen	-	51	-	-	2

45	Stereo	-	304	-	-	3
46	Orchestra	-	281	619	-	3
47	Guitar	-	795	-	-	2
48	Concert	-	895	578	386	2

#	Verb	Synonyms	Frequency*	Imageability*	Age of acquisition*	Syllables*
1	Growing	-	1527	371	-	2
2	Watering	-	109	632	153	3
3	Pruning	-	25	578	347	2
4	Fertilising	-	5	-	-	4
5	Measuring	-	126	379	344	3
6	Pouring	-	302	495	-	2
7	Tasting	-	94	425	-	2
8	Cutting	Slicing, Chopping	1126	460	-	2
9	Deciding	-	167	-	-	3
10	Buying	Purchasing	1679	397	-	2
11	Ordering	-	369	352	344	2
12	Browsing	Looking	52	-	-	2
13	Exploring	-	139	-	-	3
14	Relaxing	-	184	387	-	3
15	Packing	-	698	-	-	2
16	Negotiating	Bartering	155	-	-	5
17	Typing	-	165	395	383	2
18	Navigating	-	14	-	-	4
19	Calling	-	6311	424	225	2
20	Messaging	-	9	438	-	3
21	Volunteering	-	98	-	-	4
22	Teaching	-	1010	429	-	2
23	Socialising	-	3		-	4
24	Coaching	-	163	560	314	2
25	Helping	-	2588	464	222	2
26	Sharing	-	623	-	-	2
27	Laughing	-	2667	528	-	2
28	Hugging	-	146	-	-	2
29	Washing	-	462	522	186	2
30	Cleaning	-	1157	454	-	2
31	Multitasking	-	10	-	-	4
32	Recycling	-	80	-	-	3
33	Cheering	-	690	-	-	2
34	Handballing	-	28	-	-	3
35	Dribbling	Bouncing	31	-	-	2
36	Fishing	-	1204	615	-	2
37	Snowing	-	152	597	-	2
38	Sweating	-	419	560	-	2
39	Raining	-	499	618	211	2
40	Polluting	Contaminating	23	-	-	3
41	Exercising	-	84	-	-	4
42	Swimming	-	1019	635	256	2
43	Meditating	-	34	-	-	4
44	Hiking	-	135	-	-	2
45	Singing	-	2281	471	-	2

46	Busking	-	1	-	-	2
47	Drumming	-	57	-	-	2
48	Listening	-	3205	-	-	3

#	Adjective	Synonyms	Frequency*	Imageability*	Age of acquisition*	Syllables*
1	Unhealthy	-	83	-	-	3
2	Dirty	-	3389	485	-	2
3	Unripe	-	2	490	-	2
4	Peaceful	Tranquil	573	426	-	2
5	Sweet	-	7405	493	-	1
6	Spicy	-	169	494	-	2
7	Burnt		488	-	-	1
8	Refreshing	-	187	374	-	3
9	Poor	-	6583	447	-	1
10	Antique	-	309	549	439	2
11	Busy	-	5433	403	-	2
12	Impatient	-	206	-	-	3
13	Excited	-	2479	-	-	3
14	Frightened	Scared	1186	-	-	2
15	Multicultural	-	3	-	-	4
16	Exhausted	Tired	655	520	-	2
17	Frustrated	-	255	381	-	3
18	Broken	-	3634	469	-	2
19	Confused	Unsure	1653	-	-	2
20	Entertaining	-	288	435	-	4
21	Courageous	-	161	-	-	3
22	Friendly	-	1328	439	-	2
23	Dangerous	-	3817	-	-	3
24	Powerful	-	1791	-	-	3
25	Loud	Noisy	2031	448	-	1
26	Playful	-	59	463	-	2
27	Caring	-	359	-	-	2
28	Affectionate	-	115	471	-	4
29	Lazy	-	591	464	-	2
30	Chaotic	-	57	434	-	3
31	Heavy		2412	495	-	2
32	Overwhelmed	Stressed	201	-	-	3
33	Exhausted	Tired	655	434	-	3
34	Proud	-	4265	-	-	1
35	Competitive	Aggressive	214	-	-	4
36	Obsessed	Dedicated	526	-	-	2
37	Itchy	-	116	-	-	2
38	Freezing	Cold	680	-	-	2
39	Overcast	Cloudy	24	-	-	3
40	Mountainous	-	8	-	-	3
41	Healthy	Fit	1262	-	-	2
42	Strong	-	4430	463	-	1
43	Flexible	-	136	-	-	3

44	Challenging	Difficult	194	-	-	3
45	Classical	-	224	-	-	3
46	Famous	Talented	2296	376	-	2
47	Acoustic	Live	49	-	-	3
48	Joyful	Happy	76	473	-	2

C.2 Parameters and instruction of strategic self-cueing approach

Parameters		
Intervention targets	*Measurable aspects of functioning including aspects of body structure and function, activity, participation, and environment (Whyte et al., 2014).*	• Improved picture naming of treated items across word types (noun, verb, and adjective) at a single word level • Improved picture naming of untreated items across word types (noun, verb, and adjective) at a single word level • Improved communicative efficiency and informativeness of connected speech across discourse genres (everyday monologue, narrative monologue, and picture description)
Ingredients	*Measureable and observable actions systematically applied by the clinician toward the targets as specified above (Whyte et al., 2014).*	i. *Essential ingredients* – those which define treatment, necessary for effects to take place • Repeated practice of self-cueing strategy for skill acquisition (Maas et al., 2008) • Massed practice of self-cueing of treated items to support treatment effects (Raymer et al., 2008) • Distributed practice to enhance generalization and maintenance of strategy (Maas et al., 2008) ii. *Active ingredients* – those hypothesized to enhance the therapeutic power of treatment • Strengthening lexical-semantic, lexical-syntactic, phonological, orthographical, and autobiographical associations of treated items (Beales et al., 2016) • Self generated information for active learning (Slameka & Graf, 1978) • Use of strategy across two levels; lexical and connected speech levels (Milman, Vega-Mendoza, & Clendenen, 2014) • Training of a family member to facilitate strategy use (Simmons-Mackie et al., 2016) • Errorless learning approach to support episodic and semantic memory deficits (Jokel & Anderson, 2012) • Cognitive scaffolds to support capacity to engage in active learning (see Part 4 of Supplementary Materials) • Personal salience through the use of thematic topics and incorporation personal stimuli (Jokel et al., 2014; Kleim & Jones, 2008) iii. *Inactive ingredients* – those which do not enhance treatment effects • The nature and modality of the stimuli used to elicit lexical retrieval (e.g. black and white or colour)

Mechanism of change	*Process by which the essential and active ingredients of the treatment induce the predicted change in the treatment target (Whyte et al., 2014).*	• Stimulation – repeated exposure to treated item • Relearning – active learning of semantic, autobiographical, phonological, and orthographic information of treated words • Cognitive-relay – engagement in an internal cueing strategy • No post-intervention brain imaging was completed in the current study and therefore we are unable to ascertain the potential involvement of the reorganisation change mechanism
Dosage & timing	*Parameters of the schedule of treatment that promote and optimise functional adaptation and neural plasticity (Whyte, Gordon, & Rothi, 2009).*	*Frequency* – the number of sessions per week • 3 sessions per week (initial 2 weeks – 'massed practice') • 2 sessions per week (following 4 weeks – 'distributed practice') *Density* – the total number of minutes per session • Phase 1 and 2; 60 minutes per session • Phase 3; 90 minutes per session *Intensity* – the total number of minutes per week • 180 minutes per week (initial 2 weeks – 'massed practice') • 150 minutes per week (following 4 weeks – 'distributed practice') • Additional home practice *Duration* – the period of time between the first and last session • 6 weeks (total 14 sessions)
Instruction		
Orientation to task	*Pre-practice is essential for ensuring understanding of the problem and knowledge of performance, the goal, and the correct form for the skill or strategy to be trained (Maas et al., 2008).*	*Clear instruction of intervention target* • Explicit discussion of word finding difficulties, i.e. what are word finding difficulties? When do they happen? What kinds of errors might we make? What do we do when we experience them? • Explicit discussion of strategy use, i.e. instead of getting stuck or frustrated, what can we do to help? Are there any strategies in place already?
Direct instruction	*Comprehensive, explicit, and instructional method for teaching (Engelmann &*	*Presentation of material* • Introduce components of self-cueing with explicit reference to the self-cue support cards (e.g. meaning, memory, letter, sounds).

	Carnine, 1991).	*Clinician modelling* • Model the strategy using the three different word classes and self-cue support cards, discuss each cue and provide an example. 'Meaning' – *(semantics)* e.g. What category does it belong to? What does it look like? What is a word that means the same? When would this word be used? 'Memory' – *(autobiographical memory / salience)* e.g. Do you have one of these? Do you go to this place? When do you do this? 'Letter' – *(orthographics)* e.g. What letter does the word start with? Can you write the first letter? 'Sound' – *(phonology)* e.g. What sound does the word begin with? Can you think of other sounds in the word? • Reinforce that we are trying to work through any of the cues we might know, rather than guessing. If the participant is still unsure of the word, the clinician will prompt, provide further cue information, and then provide the target item. • To promote transference of the cueing strategy in discourse, the therapist will prompt various discourse genres. The prompts will be based on the thematic topic. The structure of discourse (macrostructure elements) will not be explicitly targeted in therapy.
Strategy-based instruction	*Teaches participates to monitor their own thinking through strategy (Swanson, 1999).*	*Metalinguistic / metacognitive teaching of self-cueing strategy* • 'Conceptualiser'- thinking of what we want to say, an intention, • 'Formulation'- accessing the meaning of the words, the sounds in the word, and the letters (if we are writing the word), arranging the words into an order (if we are putting together a phrase or sentence) • 'Articulation'- accessing the information about the movements we need to make with the muscles of our mouth to produce the sounds, • 'Cognitive'- to help find the words we want, we can also use our attention (e.g. focus, reducing distractions) and memory (e.g. of our personal experiences). *Systematic probe* • "When you can't think of a word what should you do?" Answer e.g. "Look at my cue card.

C.3 Strategic self-cue cards

Meaning
[OBJECTS/PLACES]

- Special feature – what is unique about it?
- Category – what group does it belong to?
- Visualise – what does it look like?
- Location – where is it found?
- Function – what do we use it for?
- Similar – what is it similar to?

Memory
[OBJECTS/PLACES]

- My memories – what is significant about this?
- Experiences – do you have one of these? Do you go to this place?
- Friends & family – do you know someone who has one of these? Do you know someone who goes here?

Meaning
[ACTIONS]

- Special feature – what is unique about it?
- Visualise – what does it look like?
- Location – where do we do this?
- Function – what do we need to do this?
- Similar – what is it similar to?
- Time – when do we do this?

Memory
[ACTIONS]

- My memories – what is significant about this?
- Experiences – is this something that you do? When do you do this?
- Friends & family – do you know someone who does this?

Meaning

[DESCRIBING]

- Special feature – what is unique about it?
- Visualise – what does it look like?
- Context – when would this word be used?
- Similar – what is a word that means the same?
- Opposite – what is a word that means the opposite?

Memory

[DESCRIBING]

- My memories – when do you use this word? What is the word associated with in your life?
- Experiences – describe when you last felt this way.
- Friends & family – does this word remind you of someone?

Letters

[ALL WORDS]

- First letter – what letter does it start with?
- Writing – can you write the letter?
- Letter chart – can you find the letter?
- Read – read the written word.

Sounds

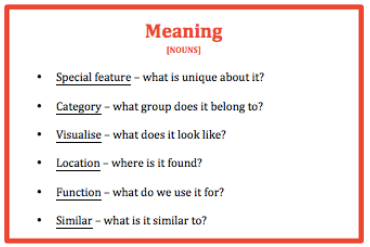
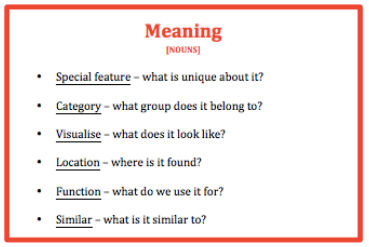
[ALL WORDS]

- First sound – what sound does it start with?
- Other sounds – can you think of any sounds in the word?
- Repetition – repeat the word.

C.4 Cognitive scaffolds implemented in intervention

Cognitive function	Scaffolds
Executive function and visual attention	• Explicit prompts to focus on use of the self-cueing strategy (i.e. increase focused attention to strategy use rather than error) • Explicit prompts to detect onset of word finding difficulty or error production (i.e. increase focused attention to communicative output) • Tasks completed in isolation, with breaks between task change (i.e. reduce impact of task shifting)
Working memory	• Instructions delivered in short phrases / units of information (i.e. minimize length of information) • Instructions delivered once the participant has completed each step in the task (i.e. minimize demand on temporary storage) • Orthographic aids to support auditory information (i.e. minimize demand on temporary storage)
Verbal episodic memory	• Increased spacing of self-cue probe "when you cannot think of a word what should you do?" (i.e. spaced retrieval principles) • Minimum of 10 exposures to the self-cue strategy each session (i.e. draw on implicit memory) • Visual aids to support task auditory information (i.e. draw on other modalities to support information processing)
Visual recognition memory	• Expose participant to one self-cue support card at a time and increase to multiple cards as able (i.e. minimize demand on processing visual stimuli) • Expose participant to one picture for treated items at a time and increase to multiple pictures as able (i.e. minimize demand on processing visual stimuli)
Autobiographical memory	• A self-cue support card for all participants to elicit personal experiences (i.e. draw on personal associations with treated items) • Inclusion of personal stimuli (e.g. photographs) in phase 3 (communication partner prompted self-cueing) to elicit meaningful discourse

C.5 Home practice instruction and record sheets

Activity	Instructions	What you need
1 **Practice pictures**	• Aim to practice for 30 minutes, 2 days per week • For each target word, try to name it. • Try not to guess the word. • If you are unsure, work through your support cards.	1. Practice pictures 2. Personal pictures 2. Support cards 
2 **Conversation**	• Aim to practice for 30 minutes, 7 days per week. • During conversation, try to use your supports when you are unsure of a word. • Your family member will help to remind you to use support cards. • Your family member will help you by providing support information that are on your cards.	1. A family member or friend 2. Support cards 

Activity 1 – Practice pictures		
Date	**Words practiced**	**Supports used**
1.		
2.		
3.		

Activity 2 – Conversation		
Conversation topics	**Words that were difficult**	**Support cues used**

C.6 Individual participant McNemar *p* values for naming performance on baseline to initial post-intervention and baseline to maintenance.

			Nouns		Verbs		Adjectives	
			Treated	Untreated	Treated	Untreated	Treated	Untreated
SvPPA	P1	Initial Post	0.035*	0.031*	0.001*	0.016*	0.008*	1.000
		Maintenance	0.008*	0.109	0.001*	0.035*	0.031*	0.125
	P2	Initial Post	0.250	0.250	0.125	0.500	1.000	0.125
		Maintenance	0.000*	0.063	0.500	0.500	0.500	0.125
	P3	Initial Post	0.063	0.063	0.016*	0.031*	0.250	0.250
		Maintenance	0.125	0.031*	0.016*	0.063	0.250	0.250
	P4	Initial Post	0.000*	0.063	0.031*	0.500	0.002*	0.125
		Maintenance	0.019*	0.188	0.035*	0.500	0.002*	0.500
LvPPA	P5	Initial Post	0.063	0.055	0.008*	0.188	0.000*	0.001*
		Maintenance	0.125	0.004*	0.016*	0.063	0.000*	0.035*
	P6	Initial Post	0.000*	0.004*	0.001*	0.000*	0.019*	0.019*
		Maintenance	0.011*	0.109	0.227	0.063	0.109	0.500
	P7	Initial Post	0.000*	0.363	0.011*	0.016*	0.031*	0.313
		Maintenance	0.000*	0.011*	0.001*	0.063	0.188	0.188
	P8	Initial Post	0.000*	0.002*	0.008*	0.001*	0.008*	0.188
		Maintenance	0.000*	0.002*	0.035*	0.019*	0.001*	0.500
AD	P9	Initial Post	0.031*	0.008*	0.019*	0.019*	1.000	0.500
		Maintenance	0.016*	0.002*	0.063	0.063	0.750	0.500
	P10	Initial Post	0.002*	0.500	0.001*	0.016*	0.016*	0.016*
		Maintenance	0.002*	0.063	0.006*	0.019*	0.016*	0.031*
	P11	Initial Post	0.109	0.063	0.008*	0.109	0.250	0.125
		Maintenance	0.500	0.109	0.031*	0.109	0.500	0.063
	P12	Initial Post	0.008*	0.313	0.125	0.063	0.063	0.250
		Maintenance	0.004*	0.109	0.145	0.227	0.125	0.313

* Significant difference ($p = < 0.05$) indicative of improved naming performance.

C.7 Individual participant Fisher Exact *p* values (z score) for %CIU on baseline to initial post-intervention and baseline to maintenance.

			Everyday Monologues	Narrative	Picture Description
SvPPA	P1	Initial Post	0.019* (2.08)	0.234 (0.73)	0.011* (2.27)
		Maintenance	0.122 (1.17)	0.225 (0.76)	0.281 (0.58)
	P2	Initial Post	0.196 (0.86)	-	0.010* (2.32)
		Maintenance	0.033* (1.84)	-	0.001* (3.56)
	P3	Initial Post	0.001** (4.24)	-	0.240 (0.71)
		Maintenance	0.121 (1.17)	-	0.286 (0.56)
	P4	Initial Post	0.375 (0.32)	-	0.001** (2.81)
		Maintenance	0.092 (1.33)	-	0.302 (0.52)
LvPPA	P5	Initial Post	0.238 (0.71)	0.035* (1.82)	0.571 (-0.18)
		Maintenance	0.298 (0.53)	0.022* (2.02)	0.287 (0.56)
	P6	Initial Post	0.444 (0.14)	-	0.099 (1.28)
		Maintenance	0.241 (0.71)	-	0.336 (0.42)
	P7	Initial Post	0.334 (0.43)	0.005** (2.55)	0.110 (1.23)
		Maintenance	0.078 (1.42)	0.332 (0.43)	0.311 (1.01)
	P8	Initial Post	0.199 (0.85)	-	0.155 (1.01)
		Maintenance	0.074 (1.45)	-	0.006** (2.51)
AD	P9	Initial Post	0.121 (1.17)	0.059 (1.57)	0.500 (0.00)
		Maintenance	0.121 (1.17)	0.241 (0.71)	0.463 (0.73)
	P10	Initial Post	0.024 (1.98)	0.332 (0.46)	0.026* (1.95)
		Maintenance	0.149 (1.04)	0.500 (0.00)	0.280 (0.58)
	P11	Initial Post	0.556 (-0.14)	0.098 (1.29)	0.556 (-0.14)
		Maintenance	0.556 (-0.14)	0.118 (1.19)	0.098 (1.29)
	P12	Initial Post	N/A	-	N/A
		Maintenance	0.174 (0.94)	-	0.361 (0.36)

* Significant one tailed ($p = < 0.05$) indicative of increase, ** significant one tailed ($p = < 0.05$) indicative of decrease, "N/A" data not available, "-" participant unable to complete assessment.

C.8 Individual participant Fisher Exact *p* values (z score) for CIUs/min on baseline to initial post-maintenance and baseline to maintenance.

			Everyday Monologues	Narrative	Picture Description
SvPPA	P1	Initial Post	0.233 (0.73)	0.245 (0.69)	0.229 (0.74)
		Maintenance	0.132 (1.12)	0.245 (0.69)	0.084 (1.38)
	P2	Initial Post	0.131 (1.21)	-	0.157 (1.01)
		Maintenance	0.235 (0.72)	-	0.157 (1.01)
	P3	Initial Post	0.010** (2.32)	-	0.401 (0.25)
		Maintenance	0.030** (1.88)	-	0.231 (0.73)
	P4	Initial Post	0.471 (0.07)	-	0.000** (3.79)
		Maintenance	0.145 (1.06)	-	0.470 (0.08)
LvPPA	P5	Initial Post	0.500 (0.00)	0.021* (2.03)	0.500 (0.00)
		Maintenance	0.290 (0.55)	0.021* (2.03)	0.237 (0.72)
	P6	Initial Post	0.418 (0.21)	-	0.313 (0.49)
		Maintenance	0.290 (0.55)	-	0.160 (0.99)
	P7	Initial Post	0.232 (0.73)	0.096 (1.21)	0.128 (1.13)
		Maintenance	0.454 (0.11)	0.537 (-0.09)	0.160 (0.99)
	P8	Initial Post	0.500 (0.00)	-	0.100 (1.31)
		Maintenance	0.299 (0.53)	-	0.000** (4.69)
AD	P9	Initial Post	0.394 (0.27)	0.289 (1.06)	0.500 (0.00)
		Maintenance	0.097 (1.30)	0.452 (0.12)	0.208 (0.81)
	P10	Initial Post	0.224 (0.76)	0.435 (0.16)	0.025* (1.95)
		Maintenance	0.608 (0.51)	0.368 (0.34)	0.104 (1.26)
	P11	Initial Post	0.298 (0.53)	0.123 (1.16)	0.475 (0.06)
		Maintenance	0.449 (0.13)	0.408 (0.23)	0.373 (0.32)
	P12	Initial Post	N/A	-	N/A
		Maintenance	0.395 (0.27)	-	0.416 (0.21)

* Significant one tailed ($p = < 0.05$) indicative of increase, ** significant one tailed ($p = < 0.05$) indicative of decrease, "N/A" data not available, "-" participant unable to complete assessment.

C.9 Statistical support for aggregated baseline naming data

All word classes

In assessing variation across the baseline period, the 3-way Condition (treated, untreated) x Diagnosis (svPPA, lvPPA, A) x Baseline Assessment (baseline 1, baseline 2, baseline 3) interaction was not significant ($F[4,48] = 1.766$, $p = .151$), and neither was the 2-way Condition x Baseline Assessment interaction ($F[2,48] = 0.921$, $p = .405$). The Diagnosis x Baseline Assessment interaction was significant, but the simple main effects of Baseline Assessment for the three diagnostic groups were all non-significant (all ps > .05).

Nouns

There was a significant 3-way Condition x Diagnosis x Baseline Assessment interaction ($F[4,48] = 4.894$, $p = .002$). In order to understand the 3-way interaction, the simple 2-way Diagnosis x Baseline Assessment interactions were subsequently analyzed for each of the two conditions. For the untreated condition, the Diagnosis x Baseline Assessment interaction was not significant ($F[4,24] = 1.621$, $p = .202$), indicating that the non-significant Baseline Assessment component ($F[2,24] = 1.299$, $p = .291$) could be generalized to all three diagnostic groups. There was, however, a significant Diagnosis x Baseline Assessment interaction for the treated condition ($F[4,24] = 15.541$, $p < .001$). The source of this interaction could be traced to a significant improvement in performance from baseline 1 to baseline 2 for the lvPPA diagnostic group ($t[24] = 8.083$, $p < .001$). Although the improvement was statistically significant, it only represented a relatively small increase (Cohen's $d = .28$), such that an aggregated baseline for nouns was used.

Verbs

The 3-way Condition x Diagnosis x Baseline Assessment interaction was not significant ($F[4,48] = 2.105$, $p = .095$), and neither was the 2-way Condition x Baseline Assessment interaction ($F[2,48] = 0.092$, $p = .912$). The Diagnosis x Baseline Assessment interaction, however, was significant ($F[4,48] = 4.242$, $p = .005$). The source of this interaction could be traced to a significant improvement in performance from baseline 1 to baseline 3 for the lvPPA diagnostic group ($t[48] = 3.266$, $p = .002$). Although the improvement was statistically significant, it only represented a relatively small increase (Cohen's $d = .24$), supporting the use of an aggregated baseline for verbs.

Adjectives

The 3-way Condition x Diagnosis x Baseline Assessment interaction was not significant ($F[4,48] = 2.353$, $p = .067$), and neither was the 2-way Condition x Baseline Assessment interaction ($F[2,48] = 1.100$, $p = .341$) nor the 2-way Diagnosis x Baseline Assessment interaction ($F[4,48] = 0.825$, $p = .516$). Because Baseline Assessment did not interact with the other factors, its non-significant main effect ($F[2,48] = 2.871$, $p = .066$) can be generalized across both conditions and all three diagnostic groups, rationalizing the use of an aggregated baseline for adjectives.

C.10 Performance means for cognitive control measures by diagnostic group and assessment time

Cognitive control	Time	svPPA	lvPPA	AD
ACE-III	Baseline	51.958	63.542	41.292
	Initial-post	53.708	61.292	42.042
Verbal episodic memory	Baseline	3.860	3.750	1.750
	Initial-post	4.271	4.673	2.035
Visual memory	Baseline	8.535	7.424	6.965
	Initial-post	6.785	8.174	6.465