The impact of Parkinson's-related dementia on life partner outcomes

A thesis submitted to the University of Manchester for the degree of Doctor of Philosophy in the Faculty of Biology, Medicine and Health

2018

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Word count: 86,085

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List of abbreviations

AD	Alzheimer's disease
AE	Adverse event
ANCOVA	Analysis of covariance
ANOVA	Analysis of variance
APIM	Actor-partner interdependence model
BRS	Brief Resilience Scale
CI	Confidence interval
CONSORT	Consolidated Standards of Reporting Trials
CST	Cognitive Stimulation Therapy
CST-PD	Cognitive Stimulation Therapy in Parkinson's-related dementia
DLB	Dementia with Lewy bodies
DRS	Dyadic Relationship Scale
EFA	Exploratory factor analysis
EPDA	European Parkinson's Disease Association
EQ-5D	EuroQoL quality of life measure
FCR	Family Caregiving Role Scale
GMMH	Greater Manchester Mental Health NHS Foundation Trust
HADS	Hospital Anxiety and Depression Scale
H&Y	Hoehn and Yahr staging system
iCST	individual Cognitive Stimulation Therapy
ISRCTN	International Standard Randomised Controlled Trial Number
ITT	Intention-to-treat
JDR	Join Dementia Research
MCAR	Missing completely at random
MCI	Mild cognitive impairment
MLM	Multilevel modelling
MoCA	Montreal Cognitive Assessment
MRC	Medical Research Council
NELFT	North East London Foundation Trust
NHS	National Health Service
NICE	National Institute for Health and Care Excellence
NIHR	National Institute for Health Research

NPI	Neuropsychiatric Inventory
NRES	National Research Ethics Service
NWBH	North West Boroughs Healthcare Trust
OECD	Organisation for Economic Co-operation and Development
ONS	Office for National Statistics
P-P plot	Probability-probability plot
PAIR	Personal Assessment of Intimacy in Relationships
PAT	Pennine Acute Trust
PD	Parkinson's disease
PDD	Parkinson's disease dementia
PD-MCI	Parkinson's disease and mild cognitive impairment
PPI	Patient and public involvement
PRD	Parkinson's-related dementia
RCT	Randomised controlled trial
REC	Research Ethics Committee
Rel.SS	Relatives' Stress Scale
RfPB	Research for Patient Benefit funding (NIHR funding)
RSS	Relationship Satisfaction Scale
SAE	Serious adverse event
SD	Standard deviation
SE-ADL	Schwab and England Activities of Daily Living Scale
SF-12	Short-Form Health Questionnaire-12
SPIRIT	Standard Protocol Items: Recommendations for Interventional Trials
SPSS	Statistical Package for Social Sciences
TAU	Treatment as usual
UHSM	University Hospital of South Manchester Trust
UK	United Kingdom
UPDRS	Unified Parkinson's Disease Rating Scale
USA	United States of America
VIF	Variance inflation factor
WHO	World Health Organisation
ZBI	Zarit Burden Interview

List of collaborators and their role

ES	Dr Emma Stanmore (Supervisor, The University of Manchester)
HM	Heena Mistry (Clinical Studies Officer, The Clinical Research Network, GMMH)
IL	Professor Iracema Leroi (Primary Supervisor, The University of Manchester)
KRM	Dr Kathryn McDonald (Supervisor, The University of Manchester)
LAC	Dr Lesley-Anne Carter (Statistician, The University of Manchester)
LC	Professor Linda Clare (Supervisor, The University of Exeter)
SAM	Dr Sheree McCormick (INVEST Trial Coordinator, The University of Manchester)
SV	Sabina Vatter (PhD Candidate, The University of Manchester)

Glossary of terms

Baseline assessments – a set of measures completed with people with Parkinson's-related dementia and life partners in an interview with blinded researchers at the start of the trial.

Blinded researchers – researchers who conducted baseline and follow-up assessments in the INVEST study and who were blinded to the randomisation allocation of participant-dyads.

Burden – the extent to which care partners perceive that their physical, emotional, psychological, social and financial status has been affected as a result of care provision. In this PhD, burden was measured with the Zarit Burden Interview (ZBI; Zarit, Reever, & Bach-Peterson, 1980).

Care partner – a person, usually a spouse, life partner or an adult child, who provides regular help and support to a family member who cannot take care of themselves due to illness, age or disability. Care partners are often referred to as informal carers or caregivers and anyone can take up this role, regardless of age or gender.

Care recipient – the person with Parkinson's-related dementia who receives regular care from a care partner (i.e. spouse or life partner).

Cognitive rehabilitation therapy – an individualised intervention which aims to support people with cognitive decline in improving their daily functioning and activities of daily living through goal-setting and implementation of rehabilitative strategies.

Cognitive stimulation therapy (CST) – a psychosocial intervention which aims to enhance cognitive and social functioning of the person with dementia or memory impairment through group activities, interactions and discussions.

Cognitive stimulation therapy in Parkinson's-related dementia (CST-PD) – an individualised home-based care partner-guided psychosocial intervention specifically adapted for people with Parkinson's-related dementia and their care partners as part of the INVEST trial. Through a therapy manual, the intervention aims to promote engagement in themed 20-30 minute conversations that stimulate thought processes, opinions, language, memory, planning and executive functioning.

Cognitive training therapy – a type of intervention which aims to enhance cognitive functioning through regular guided and repeated practice of selected standardised tasks which address specific cognitive domains, such as memory, attention, executive function, language and speed of processing. The tasks may include remembering, planning, focusing attention or organising information.

Dementia with Lewy bodies (DLB) – a neurodegenerative disorder, usually diagnosed when cognitive impairment precedes or occurs alongside the motor symptoms of

parkinsonism within one year. The most typical symptoms of DLB are cognitive impairment, fluctuating confusion, parkinsonism and visual hallucinations.

Dyad (or participant-dyads) – refers to 'care partner-care recipient' pair.

Feasibility trial – a type of clinical study design which aims to explore whether the study can be done by examining a number of study parameters required for the main randomised controlled trial (e.g. willingness of participants to be randomised, number of participants, rates of follow-up, response rates and adherence, assessing the outcome measure, etc.)

Follow-up assessments – a set of measures completed with people with Parkinson's-related dementia and life partners in an interview with blinded researchers at the end of the trial period, usually at 12 weeks.

Hoehn & Yahr (HY) staging – a scale which assesses the clinical stage of Parkinson's disease ranging from stage 1 (unilateral, one-sided symptoms) to stage 5 (most severe disease stage).

Intimate relationship – a committed long-term, cohabiting, marriage-like partnership, whereby partners may or may not be married.

INVEST trial – the overarching project name, which stands for 'INdiVidualised cognitive Stimulation Therapy'. The primary aim of the INVEST study was to adapt and trial the Parkinson's-adapted Cognitive Stimulation Therapy (CST-PD) among people with Parkinson's-related dementia and their care partners.

Life partner – a spouse or a long-term partner of a person with Parkinson's-related dementia, who is also a care partner. For clarity and consistency, all life partners who were either married or in a cohabiting relationship with the person with Parkinson's-related dementia are referred to as life partners throughout this thesis.

Parkinson's disease (PD) or Parkinson's – a complex and progressive neurodegenerative disorder characterised by motor, psychiatric and cognitive symptoms.

Parkinson's disease dementia (PDD) – is a form of dementia which develops in Parkinson's disease. It is characterised by deterioration in memory, attention, visuospatial functions, executive functions and occurrence of psychiatric symptoms, such as apathy and hallucinations. Approximately 80% of people with Parkinson's disease may be at risk of developing PDD within ten or twenty years.

Parkinson's disease with mild cognitive impairment (PD-MCI) – is a spectrum of cognitive dysfunction in Parkinson's disease. PD-MCI is characterised by deficits in at least two of the following domains: memory, language, attention and working memory, executive functions and visuospatial.

Parkinson's-related dementia (PRD) – a term referring to Parkinson's disease with mild cognitive impairment (PD-MCI), Parkinson's disease dementia (PDD) and dementia with Lewy bodies (DLB) collectively, which is used throughout this thesis. However, this is not a commonly accepted term and sometimes 'Lewy body spectrum disorders' is used instead, which also includes Parkinson's disease in addition to the three clinical syndromes.

Pilot study – a type of clinical study design which aims to undertake a smaller version of the definitive trial to test whether the components of the main study, such as recruitment, randomisation, treatment and follow-up assessments, can work together.

Randomised controlled trial (RCT) – a study in which a group of people are randomly allocated to two (or more) groups to test a specific treatment or intervention, whereby one group (the experimental group) receives the intervention and the other group (the comparison or control group) does not receive the intervention. In this PhD, participant-dyads were randomly assigned either to the CST-PD intervention group or the control group.

Relationship satisfaction – refers to one's subjective account of their level of satisfaction with the relationship. In this thesis, relationship satisfaction is measured by the seven-item Relationship Satisfaction Scale (RSS; Burns, 1983) which explores communication and openness, ability to resolve conflicts and arguments, degree of affection and caring with one's partner, intimacy and closeness, satisfaction with the respondent's role in the relationship, satisfaction with the other person's role in the relationship and overall satisfaction with the relationship.

Resilience – the ability to cope and bounce back in stressful situations.

Treatment as usual (TAU) or comparator – the standard (usual care, another intervention or no intervention) against which an intervention is compared in a trial. In this PhD, a comparator group is the control group who did not receive a supplementary intervention in addition to their standard treatment provided by the National Health Service (NHS), which included dopamine replacement therapy for the symptomatic relief of the PD symptoms, medication enhancing cognition and support from a PD nurse specialist and/or consultant. A control group allowed making comparisons between the group that received the intervention and group that did not receive the intervention.

Unblinded researchers – researchers who conducted screening and informed consent visits, randomised participant-dyads, delivered the CST-PD training to participant-dyads as well as undertook weekly phone calls in the INVEST study.

Abstract

Overall aim: To explore the impact of Parkinson's-related dementia (PRD) on the outcomes of life partners, and to investigate the effects of Parkinson's-adapted Cognitive Stimulation Therapy (CST-PD) on life partners.

Background: Care partners play a crucial role in supporting people with PRD. The literature on the impact of PRD on care partners is vast; however, less is known about the impact of PRD on life partners who take on the additional role of a care partner. As care provision takes place within dyadic relationships, the outcomes of life partners may differ compared to non-life partners. Continuous care provision in PRD can lead to adverse physical and mental health outcomes in care partners but additionally, can take a toll on relationships in life partners. Long-term relationships are important as they can determine happiness, influence physical and mental health and lengthen one's lifespan but complex neurodegenerative conditions such as PRD may significantly disrupt dyadic relationships. Caregiving experiences in PRD, with a particular emphasis on long-term relationships, were examined in this thesis from the perspective of life partners.

Methods: The objectives of the PhD were met through five related studies using different methodologies. First, with the people with PRD as the focus, the profile of life partners (n=136) was described. Second, in this same cohort, care burden was deconstructed using a factor analysis to ascertain the specific dimensions underlying the construct of burden. Third, the caregiving experiences of life partners were explored in detail using semi-structured qualitative interviews (n=12). Fourth, the associations among various health-related outcomes and relationship satisfaction were investigated in dyadic analysis with couples within PRD (n=57). Finally, the impact on life partners (n=57) of a dyadic intervention, CST-PD, was explored in a pilot randomised controlled trial.

Results: Life partners of people with PRD were mostly married women who exhibited low levels of mental health and high levels of relationship dissatisfaction, burden, stress and negative feelings, predominantly once dementia had emerged in PD. Deconstructing burden further suggested a five-factor solution, contrary to expectation. In-depth qualitative interviews revealed that life partners experienced changes in the relationship including role transitioning, multiple care-related challenges, and loss of freedom and independence due to their caregiving role. At the same time, marital vows, acceptance, adjustment and resilience were important for life partners. The dyadic analysis demonstrated that health-related outcomes and relationship satisfaction were bidirectional and the anxiety of the person with PRD impacted on relationship satisfaction of both members of the couple. Finally, CST-PD increased positive interactions with the person with PRD but changes in other domains were not noted among life partners.

Conclusions: This PhD makes a timely and valuable contribution to understanding the impact of PRD on life partners and illustrates that dyadic psychosocial interventions can be beneficial. It has important implications for future service provision for care partners. Given the benefit of including both members of the couple in the interventions, dyadic trials to support caregiving relationships, maintain quality of life and delay institutionalisation of people with PRD may be an important focus of activity in the future.

Declaration

No portion of the work referred to in this thesis has been submitted in support of an application for another degree or qualification of this or any other university or other institute of learning.

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Acknowledgments

Dedicated to my beloved grandparents who are living with neurodegenerative conditions and who keep moving forward, one step at a time, despite the challenges and setbacks ♥

With my most sincere appreciation, I would like to recognise the following individuals and teams for their support and encouragement throughout my PhD.

I am indebted to all of the participants who took part in the INVEST study, the postal survey and the qualitative interviews, this thesis would not have been possible without you. I enjoyed meeting all of you and I appreciated your time, commitment and devotion to the project. Thank you for sharing your story with me. I would also like to thank the Greater Manchester Mental Health NHS Foundation Trust for kindly sponsoring my PhD studentship.

I would like to extend my heartfelt thanks to my supervisory team, Professors Iracema Leroi and Linda Clare and Drs Kathryn McDonald and Emma Stanmore, for their invaluable comments, timely advice and continuous guidance. Ira, I am deeply grateful for your expertise, thorough feedback and suggestions, and the inspiring and thought-provoking conversations that we had. I have yet to meet someone more inspirational, ambitious and motivated than you. Katy, you have been a tremendous support throughout the writing of this thesis and I highly appreciated all your ideas, I have learnt a lot from you. Emma, your encouragement and endless positive affirmations have made my journey as a PhD student enjoyable and made me believe I am capable of achieving more. I am also thankful for the useful feedback and for bringing a different perspective to my thesis. Linda, despite the distance, you always provided thorough and insightful ideas which have helped to shape my thesis. I valued your prompt responses and expertise very much.

I cannot begin to express my gratitude to Sheree McCormick, a skilled, knowledgeable, successful and inspiring woman, who knows how to develop her ideas further, who has the courage to think outside of the box and who likes a challenge, big or small, which I genuinely admire. Through your devotion as a manager, trial coordinator and colleague, you made a fantastic team from us and thanks to you, the INVEST project was a success! You have become a good friend throughout the years and I am grateful for your time, support and guidance as a mentor, I feel privileged to know you. I value all of our fascinating and inspiring conversations and recall our attendances at the international conferences with great fondness. Thank you for taking the time to proof-read my whole thesis, I know it was not an easy task.

I would like to acknowledge all of our wonderful researchers at the sites who tirelessly worked on the INVEST project and who made a substantial contribution to the project. My gratitude is also extended to Parkinson's UK, Join Dementia Research, NHS and all the staff who helped with recruitment. Huge thanks go to all of my colleagues at the University: Jahanara, Saima, Gemma, Sophie, Emma, Zoe, Fran and Wai. I appreciated our conversations, knowledge-sharing and learning from each other and thoroughly enjoyed all of our celebrations of our successes and milestones. You are a wonderful team and I could not have asked for better colleagues.

Finally, I would like to express my deepest appreciation to my friends and family who have been there for me through my highs and lows, showed incredible support and listened to me when I needed them most. The unconditional love, patience and encouragement of my family have been unmeasurable for which I am deeply grateful. Thank you to my sister, Suzanna, who regardless of her countless queries regarding the readiness of my thesis, always showed how proud she was of me. Finally, I owe my biggest thanks to my parents, Tanja and Juri, who have always stood by my side, supported and encouraged my every decision and never stopped believing in me. Я вас очень люблю.

The Author

Sabina Vatter undertook her Bachelor of Arts (BA) and Master of Arts (MA) degrees in Psychology in Tallinn University, Estonia. She then worked at the University Hospital of South Manchester NHS Foundation Trust (now Manchester University NHS Foundation Trust) as a researcher on the 'Prevention of Delirium' study with people over the age of 65. In 2015, she commenced working on the INVEST study, where she produced a therapy manual for people with Parkinson's-related dementia and conducted a pilot randomised controlled trial of the adapted Cognitive Stimulation Therapy with people with Parkinson's-related dementia and their care partners. In 2015, she was offered a PhD sponsorship by the Greater Manchester Mental Health Trust NHS Foundation Trust.

Statement of contribution

Intervention development

Prior to commencing my doctoral studies, my role as a researcher in the INVEST study was to develop a prototype therapy manual using an iterative design process involving focus groups, individual interviews and field testing. I identified, screened and recruited participants from movement disorder clinics for the focus groups and helped to organise the focus groups. In addition, I helped to design the interview schedule and carried out individual interviews with people with Parkinson's-related dementia and care partners for the purposes of intervention development. I transcribed all focus groups and interviews and participated in the qualitative data analyses using thematic analysis. Then, I worked on the manual adaptation, finding images and photos, creating the therapy manual tasks and designing the first draft of the therapy manual between July 2015 and March 2016. I liaised with colleagues at Manchester Museum and Blackpool library to obtain images and content for the manual. I then recruited three participant-dyads where participants had a diagnosis of Parkinson's disease and mild cognitive impairment (PD-MCI), Parkinson's disease dementia (PDD) or dementia with Lewy bodies (DLB) who trialled the therapy manual. I revised the design of the manual based on the findings of the field testing stage and produced a final version ready to be trialled in the INVEST study.

INVEST pilot study

My tasks in the INVEST study were:

- Assisted in the day-to-day management of the INVEST trial and communication with participants
- Actively identified, screened, recruited and randomised participant-dyads
- Developed and implemented recruitment strategies by establishing collaborations with clinicians, Parkinson's nurses, Parkinson's UK as well as delivered talks at local Parkinson's UK meetings and Lewy Body support groups

- Undertook screening, informed consent and therapy training visits at participants' homes
- Collected and entered process evaluation data
- Conducted > 80% of weekly phone-calls with dyads in Greater Manchester and provided ongoing support to participants via telephone contacts and visits
- Edited all case reports forms
- Recorded and reported adverse events and serious adverse events
- Trained the Clinical Research Network team (i.e. blinded researchers) regarding the assessments used in the INVEST study
- Delivered protocol-specific training with the trial coordinator (S.A.M.) to the three external sites (Derbyshire, NELFT, NWBH) who joined in 2017
- Trained volunteers and research assistants who worked on the INVEST study
- Assisted with editing documents for ethics submission (including amendments)
- Participated in analysing qualitative interviews in the INVEST study
- Coordinated the data receipt from all sites
- Checked 100% of all inputted data
- Handled all data queries and research-related or therapy-related queries
- Co-wrote all INVEST manuscripts and provided feedback
- Presented INVEST findings at local, national and international conferences and meetings

PhD-specific

For my PhD, I did the following tasks:

- Undertook all 12 interviews with life partners and transcribed seven interviews
- Applied thematic analysis to analyse the qualitative data
- Conducted frequency of quotes summary
- Submitted a substantial amendment to ethics to conduct a postal questionnaire study with life partners
- Liaised with colleagues at GMMH and NWBH sites as well as Join Dementia Research and Parkinson's UK to identify life partners

- Posted out all questionnaire packs to potential participants and replied to all queries regarding the postal questionnaire study via e-mail and phone
- Inputted all postal questionnaire data
- Completed 100% of all PhD data analyses
- Participated in the 'Dementia Matters' school project, presented a talk on dementia and was filmed for the 'Dementia Matters' toolbox for teachers

List of publications

Vatter, S., McDonald, K. R., Stanmore, E., Clare, L., McCormick, S. A., & Leroi, I. (2018a). A qualitative study of female caregiving spouses' experiences of intimate relationships as cognition declines in Parkinson's disease. *Age and Ageing, 47(4)*, 604-610. [Based on research conducted in Study 3 of this thesis]

Vatter, S., McDonald, K. R., Stanmore, E., Clare, L., & Leroi, I. (2018b). Multidimensional care burden in Parkinson-related dementia. *Journal of Geriatric Psychiatry and Neurology, 31(6)*, 319-328. [Based on research conducted in Study 2 of this thesis]

Vatter, S., Stanmore, E., Clare, L., McDonald, K. R., McCormick, S., & Leroi, I. (under review). Distinct patterns of emotional, psychological and relationship factors in spouses of people with Parkinson's disease dementia or Lewy body dementia. *Journal of Geriatric Psychiatry and Neurology.* [Based on research conducted in Study 1 of this thesis]

Vatter, S., Stanmore, E., Clare, L., McDonald, K. R. & Leroi, I. Mutual influences in mental health and relationship satisfaction: a pilot dyadic analysis of couples in Parkinson's-related dementia. (In preparation for submission to a peer-reviewed journal). [Based on research conducted in Study 4 of this thesis]

Other relevant publications to this thesis

McCormick, S. A., McDonald, K. R., **Vatter, S.,** Orgeta, V., Poliakoff, E., Smith, S. J., & Leroi, I. (2017a). Psychosocial therapy for Parkinson's-related dementia: intervention development. *Clinical Interventions in Aging, 12*, 1779-1789.

McCormick, S. A., McDonald, K. R., **Vatter, S.,** Orgeta, V., Poliakoff, E., Smith, S. J., Silverdale, M. A., Fu, B., & Leroi, I. (2017b). Psychosocial therapy for Parkinson's-related dementia: study protocol for the INVEST randomised controlled trial. *BMJ Open, 7(6)*, e016801.

McCormick, S. A., **Vatter, S**., Carter, L.-A., Smith, S. J., Orgeta, V., … Leroi, I. (in press). Parkinson's-adapted Cognitive Stimulation Therapy: Feasibility and acceptability in Lewy body spectrum disorders. *Journal of Neurology*

Leroi, I., **Vatter, S**., Carter, L.-A., Smith, S. J., Orgeta, V., … McCormick, I. (under review). Parkinson's-adapted Cognitive Stimulation Therapy: A pilot randomised controlled clinical trial. *Therapeutic Advances in Neurological Disorders.* [Based, in part, on research conducted within Study 5 of this thesis]

List of presentations

Vatter, S., McDonald, K., Poliakoff, E., Smith, S., Orgeta, V., & Leroi, I. (29th of January, 2016). Poster presentation entitled *"Adaptation of a psychosocial therapy for people with Parkinsonian dementia"* at the **British Geriatrics Society BritMODIS Conference**, Birmingham, UK *(best poster award).*

Vatter, S. (2nd and 11th of February, 2016). Oral presentation of the INVEST study to two schools in Greater Manchester as part of the **Dementia Matters school project**, which aimed to increase knowledge about dementia among young people.

Vatter, S. (30th of June, 2016). Image presentation entitled *"Fun-dementially changing lives in Parkinsonian dementia"* at the **University of Manchester's Postgraduate Summer Research Showcase.**

Vatter, S., McDonald, K., McCormick, S., & Leroi, I. (7th of November, 2016). Poster presentation entitled "*What impacts relationship satisfaction in Parkinsonian dementias?"* at the **Parkinson's UK Annual Conference**, Leeds, UK.

Vatter, S., McDonald, K., McCormick, S., & Leroi, I. (9th of January, 2017). Poster presentation entitled "*What impacts relationship satisfaction in Parkinsonian dementias?"* at the **University of Manchester's School of Biological Sciences launch** *(runner-up prize).*

Vatter, S. (16th of May, 2017). Oral presentation entitled *"Changing patterns of intimate relationships as dementia emerges in Parkinson's disease"* and Poster presentation entitled "*What impacts relationship satisfaction in Parkinsonian dementias?"* at the **University of Manchester's Doctoral Academy Conference.**

Vatter, S., McDonald, K., Stanmore, E., McCormick, S., & Leroi, I. (5th of June, 2017). Poster presentation entitled *"Changing patterns of intimate relationships as dementia emerges in Parkinson's disease"* at the **Movement Disorder Society's Annual Congress**, Vancouver, Canada.

Vatter, S., McDonald, K., Stanmore, E., McCormick, S., & Leroi, I. (5th of June, 2017). Poster presentation entitled *"What impacts relationship satisfaction in Parkinsonian dementias?"* at the **Movement Disorder Society Annual Congress**, Vancouver, Canada.

Vatter, S., McDonald, K., McCormick, S., & Leroi, I. (4th of July, 2017). Oral presentation entitled *"Changing patterns of intimate relationships as dementia emerges in Parkinson's disease"* at the **British Society of Gerontology Annual Conference,** Swansea, UK.

Vatter, S. (21st of February, 2018). Oral presentation (three-part symposium) entitled *"Care partners of people with Parkinson's-related dementia"* at the **Institute for Collaborative Research on Ageing (MICRA) seminar, University of Manchester.**

Vatter, S., Stanmore, E., McDonald, K., McCormick, S., Clare, L., & Leroi, I. (17th of March, 2018). Poster presentation entitled *"The profile and burden of caregiving life partners of people with Parkinson's-related dementia*" at the **American Association for Geriatric Psychiatry Annual Conference,** Honolulu, Hawai'i, USA.

Vatter, S., Stanmore, E., McDonald, K., McCormick, S., Clare, L., & Leroi, I. (4th of July, 2018). Poster presentation entitled *"Relationship dissatisfaction in caregiving spouses of people with Parkinson's-related dementia*" at the **British Society of Gerontology Annual Conference,** Manchester, UK.

Vatter, S. (5th of July, 2018). Oral presentation (four-part symposium) entitled "*The role of the care partner in Parkinson's-related dementia outcomes*" at the **British Society of Gerontology Annual Conference,** Manchester, UK.

Ethical approval and trial registration

The INVEST study was funded by the National Institute of Health Research (NIHR), Research for Patient Benefit (RfPB) grant scheme (competition number 22, grant number: PB-PG-0613-31058) and registered with ISRCTN (registration number: ISRCTN11455062). Ethical approval for the study was granted by the Yorkshire & the Humber – Bradford Leeds (reference number: 15/YH/0531) in January 2016 (Appendix A).

The trial was registered with the following sites:

- Greater Manchester Mental Health NHS Foundation Trust (GMMH)
- Salford Royal NHS Foundation Trust (SRFT)
- Pennine Acute Hospitals NHS Trust (PAT)
- University Hospital of South Manchester NHS Foundation Trust (UHSM)
- Derbyshire Healthcare NHS Foundation Trust (Derbyshire)
- North East London NHS Foundation Trust (NELFT)
- North West Boroughs Healthcare NHS Foundation Trust (NWBH)

CHAPTER 1: Introduction

1.1 Introduction

Neurodegenerative conditions such as Parkinson's disease (PD), Alzheimer's disease (AD) and dementia with Lewy bodies (DLB) are growing in prevalence due to an ageing population. Worldwide, approximately 10 million people are affected by PD, of whom 145,500 people live in the UK. PD is one of the fastest growing neurological conditions and has recently been named the 'Parkinson pandemic'. A large proportion of people with PD will develop mild cognitive impairment (PD-MCI) or dementia (PDD) within twenty years of being diagnosed with PD, which significantly impacts people with PD and care partners and has implications for healthcare system. Cognitive impairment and dementia in PD (collectively referred to as 'Parkinson's-related dementia', PRD, throughout this thesis) are the leading cause of institutionalisation, and increases in healthcare costs and mortality. However, due to the care and support provided by care partners, significant savings in the healthcare economy, which annually exceeds £11.6 billion in the UK, can be realised. This underscores the importance of the help that care partners provide.

As PD progresses and cognitive impairment emerges, the help required by people with PD increases significantly. Frequently, the role of care partners is fulfilled by spouses and life partners, who help to manage symptoms, provide continued assistance and arrange care. However, care partners have to make social, financial and personal adjustments in order to accept and embrace their new role. As a result, care partners can feel overwhelmed, burdened, stressed and have poor mental health. To date, the majority of the studies on informal caregiving in the field of dementia have focused on examining the impact of Alzheimer's disease (AD) on care partners and have often excluded care partners of other types of dementia. Thus, it is crucial to explore the impact of lesser known dementias, such as PDD and DLB, on care partners in order to understand the extent of the impact on care partners.

Several pharmacological and non-pharmacological treatment options addressing motor and psychiatric symptoms are available for people with PD but often these may not be suitable for people with a complex type of dementia, such as PDD and DLB. Cognitive impairment has been found to be the main factor contributing and predicting negative outcomes for both people with PD and their care partners; thus, it is important to target dementia-specific outcomes, such as changes to behavioural symptoms, quality of life and social interactions with others. One option is to develop dyadic care partner-guided psychosocial interventions that are tailored to people with PRD. However, studies that have explored the efficacy, tolerability and cost-effectiveness of these approaches are lacking in PRD. Research conducted outside of PDD and DLB demonstrated that psychosocial interventions can be efficacious in improving quality of life in people with dementia and relationship quality in care partners, which were similar to the areas that people with dementia wished to gain from psychosocial interventions. This suggests there is scope for psychosocial interventions specifically adapted for people with PRD and their care partners that could potentially bring similar benefits.

In light of the Prime Minister's Dementia Challenge 2020 aiming to make England the leading country in the world for dementia care, support and research, it is important and timely to develop and trial Parkinson's-specific psychosocial interventions. This urgency led to the INVEST study, a pilot feasibility randomised controlled trial (RCT) of Parkinson's-adapted Cognitive Stimulation Therapy (CST-PD) for people with PRD and their care partners. The question remains, however, whether this type of intervention can bring meaningful benefits for caregiving life partners, resulting in positive and long-lasting effects for couples.

1.2 Overview of the thesis

This thesis is divided into four parts: (1) an overview of the theoretical background of PRD, care provision in PRD and intimate relationships in PRD (Chapter 2); (2) a detailed exploration of the impact of PRD on the outcomes of life partners using quantitative and qualitative methods (Chapters 5-8); (3) a pilot exploration of the impact of an

adapted Cognitive Stimulation Therapy in Parkinson's-related dementia (CST-PD) on life partners (Chapter 9); and, (4) a final summing up of the study findings, including critical appraisal (Chapter 10) (see Figure 1.1).

Chapter 2 provides a thorough overview of PD and cognitive impairment in PD, treatment opportunities, impact of PRD on life partners and intimate relationships as well as psychosocial interventions for people with PRD and life partners.

Chapter 3 introduces the research aims of the thesis.

Chapter 4 describes the quantitative methods of the PhD studies in detail.

Chapter 5 outlines a cross-sectional study of life partners of people with PRD and describes their sociodemographic and clinical syndrome. Additionally, comparisons of the outcomes of life partners according to the diagnosis type of the care recipient and evaluation of the psychometric properties of the scales are provided in this chapter.

Chapter 6 examines the factor structure of the Zarit Burden Interview in order to understand the unique aspects of how burden is experienced among life partners of people with PRD and to build on previous findings. This chapter also describes the associations and predictors among the clinical variables and the factors that emerged from the analyses.

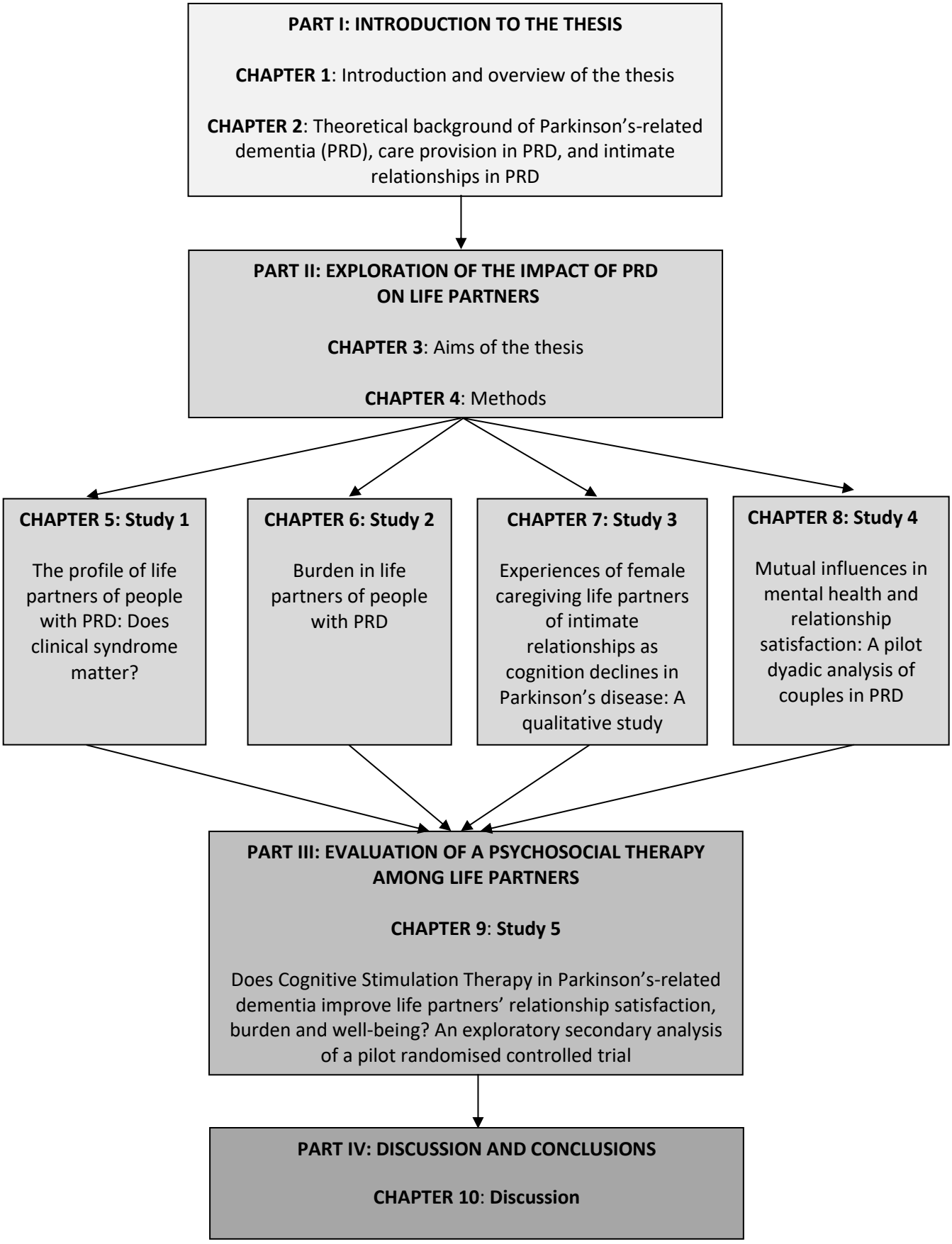
Chapter 7 outlines a qualitative study involving semi-structured interviews which explore in-depth changes to the long-term relationships and intimacy of life partners of people with PRD and how they have adjusted to the changes.

Chapter 8 describes an actor-partner interdependence model which was used to analyse the interactions between depression, anxiety, quality of life and relationship satisfaction among people with PRD and their life partners. Specifically, this chapter describes the actor and partner effects of the studied variables.

Chapter 9 draws on the previous study chapters and outlines an initial analysis of a pilot randomised controlled trial of CST-PD to explore whether intrapersonal and interpersonal aspects of life partners, such as relationship satisfaction, mental health, quality of life and burden, improve as a result of participating in a 12-week CST-PD intervention.

Chapter 10 concludes this thesis by summarising the findings of the studies and discussing them in the context of previous research and providing a critical analysis of the work. This chapter also provides implications for healthcare and makes recommendations for future research.

Figure 1.1 The structure of the thesis.



CHAPTER 2: Theoretical background

2.1 Overview of Parkinson's-related dementia (PRD)

> *"If you asked my kids to describe me, they'd go through a whole list of words before even thinking about Parkinson's. And honestly, I don't think about it that much either. I talk about it because it's there, but it's not my totality."*
>
> *- Michael J. Fox*

2.1.1 Parkinson's disease (PD)

Parkinson's disease (PD) is a complex progressive movement disorder characterised by multiple motor and non-motor symptoms and first described in 1817 by James Parkinson (Parkinson, 2002). PD affects about 10 million people worldwide and is the second most common neurodegenerative condition after AD (EPDA 2018). In 2018, it is estimated that 145,500 people have been diagnosed with PD in the UK and this figure is expected to rise to 168,000 by 2025 (Parkinson's UK, 2017a). PD is 1.5 times more prevalent for men aged 50-89 than for women in the same age range (Parkinson's UK, 2017a). A recent 'Global Burden of Disease Study' concluded that PD is one of the most rapidly growing neurological conditions for which the number of deaths, prevalent cases and disability-adjusted life years have doubled between 1990 and 2015 (GBD 2015 Neurological Disorders Collaborator Group, 2017). This has been termed the 'Parkinson Pandemic' (Dorsey & Bloem, 2018).

To diagnose PD, primary motor symptoms including slowness of movement (i.e. bradykinesia) accompanied by muscular rigidity, rest tremor, or postural instability should be present (Appendix B, Table B.1; Gibb & Lees, 1988). Bradykinesia, often one of the first presenting symptoms, is the slowed initiation of a voluntary movement, where speed and amplitude of repetitive actions is reduced (Gibb & Lees, 1988; Hughes, Daniel, Kilford, & Lees, 1992). The most common secondary motor symptoms

may include hypomimia (masked face), dysarthria (motor speech disorder), dysphagia (difficulty in swallowing), sialorrhea (hypersalivation or drooling), micrographia (abnormally small handwriting), shuffling gait, festination (quickening of gait), freezing (sudden stopping of movement) and dystonia (uncontrollable muscle contraction or spasm) (Jankovic, 2008). It has now been widely recognised that PD does not only manifest physical symptoms but also includes a myriad of non-motor symptoms such as neuropsychiatric and cognitive abnormalities, autonomic dysfunction, sleep disturbances and sensory abnormalities (Chaudhuri, Healy, & Schapira, 2006; Chaudhuri, Odin, Antonini, & Martinez-Martin, 2011; Jankovic, 2008; Appendix B, Table B.1). Indeed, Langston (2006) concluded that parkinsonism is just the 'tip of the iceberg' and should rather be seen as the 'Parkinson's complex' due to the motor and non-motor symptoms.

PD requires a thorough clinical assessment on the grounds of the complex and multifaceted nature of the illness. Typically, the motor symptoms are assessed using a five-step staging system called the Hoehn & Yahr scale (H&Y; Hoehn & Yahr, 1967) and a more detailed motor examination sub-scale called the Unified Parkinson's Disease Rating Scale (UPDRS-III; Goetz et al., 2008a). The H&Y scale describes PD severity from Stage 1 (unilateral) to Stage 5 (debilitation and confinement). The H&Y scale has been in use for over half a century; however, a modified version of H&Y is preferred by movement disorder specialists (Goetz et al., 2004), which includes two additional stages; both versions are presented in Table 2.1. The UPDRS-III specifically examines the motor symptoms of PD, including speech, facial expression, rigidity, tremor, finger and hand movements, leg raising ability, arising from chair, posture, body bradykinesia, gait and freezing of gait (Goetz et al., 2007), thus providing a more individualised description of PD motor symptoms than the H&Y scale. A range of scales have also been developed to measure the non-motor symptoms of PD, for example the Non-Motor Symptom Assessment Scale for Parkinson's disease (Chaudhuri et al., 2006), but a significantly larger body of literature has specifically focused on assessing the psychiatric and cognitive manifestations of PD through self-, informant- and/or clinician-rated scales. Despite the existing motor examination scales, diagnosing PD remains a challenge with many cases being misdiagnosed or underdiagnosed due to

the high variability and individuality of symptom presentation between people with the condition. This argues for a comprehensive clinical assessment for each individual (Jankovic, 2008) and the close monitoring of presenting symptoms over time.

Table 2.1 The Hoehn & Yahr Scale (Hoehn & Yahr, 1967) and the modified Hoehn & Yahr Scale (Goetz et al., 2004).

Hoehn & Yahr Scale	Modified Hoehn & Yahr Scale
0 No signs of disease	**0** No signs of disease
1 Unilateral involvement only, usually with minimal or no functional disability	**1.0** Unilateral involvement only **1.5** Unilateral and axial involvement
2 Bilateral or midline involvement without impairment of balance	**2.0** Bilateral involvement without impairment of balance **2.5** Mild bilateral disease with recovery on pull test
3 Bilateral disease: mild to moderate disability with impaired postural reflexes; physically independent	**3.0** Mild to moderate bilateral disease; some postural instability; physically independent
4 Severely disabling disease; still able to walk or stand unassisted	**4.0** Severe disability; still able to walk or stand unassisted
5 Confinement to bed or wheelchair unless aided	**5.0** Wheelchair-bound or bedridden unless aided

2.1.2 Cognitive impairment in Parkinson's disease

Three cognitive dysfunction stages have been posited in PD: (1) no cognitive impairment, which may include subjective cognitive impairment, (2) PD with mild cognitive impairment (PD-MCI) and (3) Parkinson's disease dementia (PDD). Additionally, if cognitive symptoms predate motor symptoms, or occur within the first twelve months following the onset of motor symptoms, dementia with Lewy bodies (DLB) may be diagnosed (Mrak & Griffin, 2007). PD-MCI, PDD and DLB are jointly referred to as 'Lewy body spectrum disorders' (Aarsland, 2016; Goldman, Williams-Gray, Barker, Duda, & Galvin, 2014; Zweig & Galvin, 2014). Specific guidelines have been developed for diagnosing each of the clinical profiles, as outlined in Appendix B (Table B.1). To facilitate referring to PD-MCI, PDD and DLB collectively, the term Parkinson's-related dementia (PRD) was chosen by the author and is used throughout this thesis.

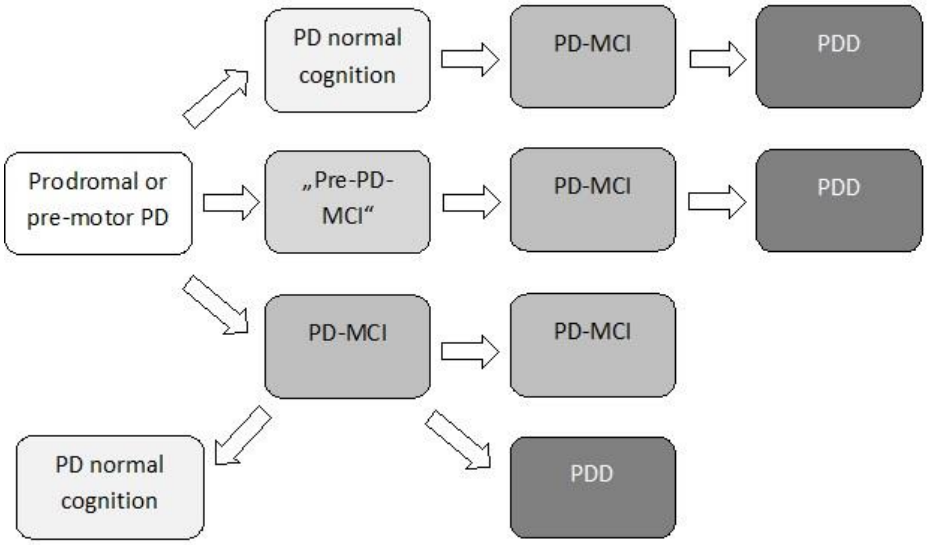
2.1.2.1 PD with mild cognitive impairment (PD-MCI)

Mild cognitive impairment (MCI) represents a change in cognition, which sits between normal cognitive functioning and early dementia (Petersen, 2004). MCI (extraneous to movement disorders) is diagnosed in the presence of a subjective memory complaint (preferably corroborated by an informant), an objective memory impairment relative to age and education, normal general cognitive function, intact or minimal decline in functional activities of daily living, and absence of dementia (Petersen, 2004). Amnestic-type MCI, where memory loss is predominant, has a high risk of transforming into AD, whereas non-amnestic MCI, where impairments occur in domains other than memory, is more likely to convert into other types of dementia such as frontotemporal dementia, vascular dementia or dementia with Lewy bodies.

Over the last two decades, researchers have increasingly recognised that cognitive impairment may occur during early stages of PD. In fact, nearly 25% of newly diagnosed people with PD without dementia have been found to present with cognitive impairment (Muslimovic, Post, Speelman, & Schmand, 2005). This finding has been confirmed by other studies (Aarsland, Bronnick, Larsen, Tysnes, & Alves, 2009; Foltynie, Brayne, Robbins, & Barker, 2004). A multicentre pooled study including 1346 people with PD concluded that 26% of subjects had MCI (range 19%-39%) and memory impairment was the most common form of cognitive decline (13.3%) (Aarsland et al., 2010). PD-MCI is associated with older age, male gender, and depression as well as duration and severity of PD (Aarsland et al., 2010; Litvan et al., 2011). PD-MCI is also known as a well-established precursor to dementia (Hobson & Meara, 2015; Litvan et al., 2012). The most common type of cognitive impairment in PD-MCI is non-amnestic followed by the amnestic type (Aarsland et al., 2010). Goldman and colleagues (2018a) have proposed that PD-MCI can remain as PD-MCI, progress to PDD or revert back to normal cognition (see Figure 2.1). The authors also argue that a stage of 'pre-PD-MCI' can become more common, particularly if an individual with PD is experiencing some cognitive symptoms that do not yet meet diagnostic criteria for PD-MCI (Goldman et al., 2018a). A recent study evaluated two groups of people with PD, those who had PD-MCI and those who were cognitively intact (Jones, Kuhn, & Szymkowicz, 2018). The

study found that people with PD-MCI who reverted to normal cognition had a higher risk of developing PD-MCI or PDD in the future at the second, third and fourth annual follow-up compared to those people that remained cognitively intact over four years (Jones et al., 2018). This suggests that cognitive impairment is central in the development of PD-MCI.

Figure 2.1 Potential cognitive impairment trajectories in PD (Goldman et al., 2018a; reproduced with permission from John Wiley and Sons, license number: 4457041016911).



2.1.2.2 Parkinson's disease dementia (PDD)

Dementia in PD (PDD) has become increasingly prevalent with nearly 80% of people with PD developing dementia within a decade (Aarsland, Andersen, Larsen, Lolk, & Kragh-Sørensen, 2003) or two decades (Hely, Reid, Adena, Halliday, & Morris, 2008) after receiving the diagnosis of PD. PDD is characterised by deterioration in memory, attention, visuospatial functions, executive functions and occurrence of behavioural and psychiatric symptoms, such as apathy and hallucinations (Emre et al., 2007; Goetz, Emre, & Dubois, 2008b). Of these presenting symptoms, having a limited cognitive reserve, MCI at baseline (Emre et al., 2007) and hallucinations (Aarsland et al., 2003) are the main risk factors for developing PDD, alongside with older age, more severe

PD, predominant gait dysfunction and older age at diagnosis (Emre et al., 2007; Riedel et al., 2008). Each year about 11% of people with PD develop dementia (Hobson & Meara, 2015), which could triple by 2060 (Savica, Grossardt, Rocca, & Bower, 2018), highlighting the urgent need to focus on this population. Furthermore, by the time PD-MCI is diagnosed, the chances of advancing to PDD are fourfold compared to PD without cognitive impairment (Hobson & Meara, 2015), indicating that once cognition has started to decline in PD, the diagnosis of dementia will be likely.

2.1.2.3 Dementia with Lewy bodies (DLB)

Dementia with Lewy bodies (DLB) is considered to be the second most common type of neurodegenerative dementia following AD (Barker et al., 2002) with a prevalence of 4.2-4.6% of all dementia cases (Kane et al., 2018; Vann Jones & O'Brien, 2014). Pathologically, the distinctive feature of DLB is the appearance of the thread-like protein deposits containing pathologic alpha-synuclein (known as the Lewy bodies) which occur in the central, peripheral, and autonomic nervous system (Jellinger, 2009; Mueller, Ballard, Corbett, & Aarsland, 2017). Most often, the symptoms of DLB include cognitive impairment (especially in visuospatial domains and executive function), fluctuating confusion, parkinsonism, visual hallucinations, sleep disturbances and apathy (McKeith et al., 2017; Mueller et al., 2017). Unlike PDD, which is diagnosed within firmly established PD, the clinical diagnosis of DLB is diagnosed when cognitive impairment precedes or occurs alongside the motor symptoms of parkinsonism (McKeith et al., 2017). Recent evidence suggests that 'pure DLB' is less common than 'DLB with concurrent Alzheimer's pathology' due to the overlap of Lewy bodies and neurofibrillary tangles specific to AD (Barker et al., 2002; Mrak & Griffin, 2007; Mueller et al., 2017). However, comparative studies have demonstrated that cognitive decline is faster in DLB than in AD (Rongve et al., 2016) confirming that DLB is an independently standing disease entity.

In 2017, the international clinical criteria for diagnosing and managing DLB were updated and now include guidelines for differentiating between clinical features and diagnostic biomarkers (McKeith et al., 2017; Appendix B, Table B.1). However,

screening of people with DLB in the studies of this PhD were undertaken according to the previous criteria of diagnosing DLB which do not include biomarkers (McKeith et al., 2005).

2.1.2.4 Comparison of PDD and DLB

Many researchers have debated whether PDD and DLB are the same disease or not due to overlap in cognitive, motor and neuropsychiatric features (Boeve et al., 2016; Friedman, 2018; McKeith et al., 2017; Taylor & O'Brien, 2012). Likewise, the one-year window, which differentiates between PDD and DLB, has been found to be arbitrary as there is no strong pathological or clinical evidence to demonstrate its validity (Taylor & O'Brien, 2012). Some researchers have concluded that PDD and DLB do not differ in regards to cognitive, attentional and neuropsychiatric profiles, sleep and autonomic dysfunction, PD type and severity, neuroleptic sensitivity, and responsivity to cholinesterase inhibitors (Aldridge, Birnschein, Denburg, & Narayanan, 2018; Friedman, 2018; Jellinger & Korczyn, 2018; McKeith et al., 2004; McShane, 2008; Noe et al., 2004; Thomas et al., 2005). However, this has been challenged by further studies undertaken with both populations (see Table 2.2). For example, significant differences have been described for age of onset (PDD<DLB; Fields, 2017), levodopa responsiveness (DLB<PDD; Dodel et al., 2008) and neuropsychological test performance (DLB<PDD; Mondon et al., 2007). Furthermore, a comparative study including people with PDD, DLB and AD concluded that the neuropsychiatric symptom presentation in PDD is more similar to AD, than to DLB (Chiu, Tsai, Chen, Chen, & Lai, 2016), suggesting that PDD and DLB are separate clinical conditions but share a common underlying pathology. Despite the contradictory findings about similarities and differences between PDD and DLB, it has been recommended that in making a clinical diagnosis, the twelve-month rule separating DLB diagnosis from PDD should be followed due to its convenience (McKeith et al., 2017) and that clinicians should verify the diagnosis via individual clinical assessment of each person.

Table 2.2 Differentiating PDD and DLB (Dodel et al., 2008; Fields, 2017; McShane, 2008; Wand, 2007).

Features:	PDD	DLB
Age of onset	60-70	65-75
Gender	More common in men	More common in men
Dementia onset in regards to PD	Later	Earlier (65-75)
Dementia progression	More rapid compared to PD, dementia increases mortality	Dementia at diagnosis, either rapid (1-5 years) or moderate annual decline
PD motor symptoms	100% More asymmetrical	25-50%, less tremor More symmetrical
Average survival following diagnosis	12-20 years	6.5-7.5 years
Motor and non-motor symptom progression	Slower	Faster
Cognitive functioning & fluctuations (e.g. attention)	Moderate to severe	Severe
Memory deficits	Deficit in recall, but not learning	Recall and learning
Psychiatric symptoms (e.g. visual hallucinations, delusions)	Common, onset often after levodopa	Very common, spontaneous onset, often at presentation
Other neuropsychiatric symptoms (e.g. depression, apathy, anxiety)	Common	Common
Rapid eye movement sleep behaviour disorder	Common	Common
Response to levodopa	Common	Variable

Abbreviations: DLB – dementia with Lewy bodies; PD – Parkinson's disease; PDD – Parkinson's disease dementia.

2.1.3 Impact of Parkinson's-related dementia

Both PD and PRD have a significant impact on the person with the condition, their life partner, and family, as well as on society, due to higher needs and dependency as a result of developing the illness. For the person with the condition, the progression of PD can worsen their health-related quality (Vescovelli, Sarti, & Ruini, 2018), particularly physical and social functioning, cognition, communication and emotional well-being (Schrag, Jahanshahi, & Quinn, 2000). The notion of adverse impact of PD on physical, social and role functioning is corroborated by a qualitative study which found that PD brings about many changes in emotions and feelings, including fears and uncertainty about the future but also highlights some benefits that PD may bring (Chiong-Rivero et al., 2011). Despite the well-established association between subjective well-being and motor impairment, there is a growing literature suggesting

that more emphasis should be paid to the positive aspects of well-being, specifically endorsing social support, socialising with other people with PD, engaging in physical activities and maintaining motor skills can contribute to life satisfaction, sense of accomplishment, autonomy and positive emotions in people with PD (Vescovelli et al., 2018). This suggests that future studies could focus on life satisfaction and psychological well-being, which could potentially diminish the negative impact of PD on the person.

In terms of the wider impact of PD on society, the disease places a major socio-economic burden with an estimated annual cost of £2 billion in the UK (Wong, 2013). A recent report on the impact of living with PD revealed that the total financial costs per household exceeded £16,000 per year due to increase in health and social care costs and reduction in income (Gumber et al., 2017). McLaughlin and colleagues (2010) found that many care partners of people with PD had to give up employment to be able to provide care for their partner, which led to loss of income. As the severity of PD increases, the costs also rise and can be up to six times higher at the advanced stage (i.e. H&Y stage 5) compared to the initial stage (H&Y stage 1) (Findley, 2007). These costs likely increase with disease progression due to the complexity of concomitant symptoms of PD, the increasing need for a care partner, and increased rate of admission to residential care homes. However, some of the costs could be partially saved by the help, care and support that family care partners provide to people with PD and PRD. Prince and colleagues (2014) estimated that care providers save about £11.6 billion in the UK each year, which is increasing faster than the corresponding increase in formal health and social care costs (Prince et al., 2014).

Cognitive impairment in PD significantly increases the frequency of institutionalisation (Aarsland, Larsen, Tandberg, & Laake, 2000; Safarpour et al., 2015) and increases healthcare costs even more than PD without cognitive impairment (Hughes, Ross, Mindham, & Spokes, 2004; Larsson, Torisson, & Londos, 2018). Furthermore, mortality, which is already increased among people with PD compared to the rest of the population (Hobson & Meara, 2018), increases with the emergence of dementia, which is one of the key predictors of PD-related mortality (Hughes et al., 2004; Larsson,

Torisson, & Londos, 2018). The emergence of cognitive impairment can also significantly decrease quality of life of people with PD and increase emotional stress (Lawson, Collerton, Taylor, Burn, & Brittain, 2018).

Similarly to PD and PDD, a diagnosis of DLB can also escalate healthcare costs (Zweig & Galvin, 2014), shorten time to death (Oesterhus et al., 2014; Price et al., 2017; Zweig & Galvin, 2014), and accelerate the rate of admission to residential care homes and hospitals (Rongve, Vossius, Nore, Testad, & Aarsland, 2014; Zweig & Galvin, 2014). A DLB diagnosis can also lengthen hospital stay and increase hospitalisation costs (Mueller et al., 2018; Vossius et al., 2014) compared to AD. Mueller *et al.* (2018) explain that this is due to deteriorated physical health and increased neuropsychiatric symptoms in DLB and they conclude that overall people with DLB have a worse prognosis compared to people with AD (Mueller et al., 2017). Mueller and colleagues (2018) estimate that approximately 80,000 people with DLB in the UK will incur over 27,000 hospital admissions, and spend over 300,000 days in hospital that will exceed £35 million in hospitalisation costs in just one year, which is higher compared to the equal number of people with AD.

The aforementioned findings demonstrate that cognitive impairment and dementia in the context of parkinsonian disorders have a substantial impact on the society due to the increased risk of admissions to hospitals and residential care homes and due to the associated healthcare costs. Given that the costs will keep rising due to the ageing population and higher proportion of people developing dementia in the future, it is crucial to develop cost-effective interventions for people with PRD and their care partners that help to maintain quality of life, preserve couples' relationships and thus delay institutionalisation.

2.1.4 Pharmacological treatment of Parkinson's-related dementia

Taking into account the complexity and multidimensionality of PD, individual assessment of each person's needs, symptoms and comorbidities is required to prescribe an appropriate treatment plan and medication regime to ensure high-quality

care provision for people with PD [National Institute for Health and Care Excellence, (NICE), 2018a]. One of the most effective PD medications is levodopa which is a natural amino acid that converts into dopamine; in turn, it significantly reduces stiffness and bradykinesia (Parkinson's UK, 2015). Nonetheless, after several years of levodopa therapy, the effects of the drug can wear off causing side effects such as dyskinesia (involuntary movements), 'on-off' periods when the treatment unexpectedly stops or starts working, and a higher frequency of bradykinesia, tremor and rigidity prior to receiving the next dose (Reichmann, 2016). Other medications include dopamine agonists, monoamine oxidase type B inhibitors (MAO-B), catechol-O-methyltransferase inhibitors (COMT), glutamate antagonists (i.e. Amantadine) and anticholinergics, which are prescribed alongside levodopa or separately (Parkinson's UK, 2015). Surgery can be occasionally recommended when medication does not prove effective (e.g. for dyskinesias) and includes lesioning (damaging or ablating certain cells in the brain) or deep brain stimulation (fine wires with electrodes are entered into specific parts of the brain) (Parkinson's UK, 2017b). On the 200th anniversary of Dr James Parkinson's discovery of the 'shaking palsy' (which today is what we know as Parkinson's disease) movement disorder specialists published a summary of the past, present and future of PD stating that disease modification continues to be the main priority in the future for the PD treatment (Obeso et al., 2017).

For the treatment of cognitive impairment related to neurodegenerative parkinsonism, specifically PDD and DLB, several medications are available. The findings from meta-analyses have determined that cholinesterase inhibitors (i.e. rivastigmine, donepezil) are beneficial in terms of cognition, behavioural disturbances and global functioning for people with PDD and DLB (Matsunaga, Kishi, Yasue, & Iwata, 2016; Rolinski, Fox, Maidment, & McShane, 2012; Stinton et al., 2015). Memantine, a N-methyl D-aspartate receptor antagonist, could also improve global clinical status and neuropsychiatric symptoms in people with mild to moderate DLB but not in people with PDD (Emre et al., 2010). However, when people with PDD were taking memantine, the burden among care partners may be reduced (Leroi, Atkinson, & Overshott, 2014). Drug-based treatments are often the first suggested treatment choice for many people with PD, PDD and DLB due to their evidence-base and efficacy;

however, medication and surgeries can be expensive, may not suit each individual, have modest effects, cause side effects and can even result in worsening of motor and cognitive symptoms in PD (Walton, Naismith, Lampit, Mowszowski, & Lewis, 2017; Xie, Meng, Xiao, Zhang, & Zhang, 2016; Yang, Sajatovic, & Walter, 2012). Therefore, looking beyond pharmacological treatment is necessary.

2.1.5 Non-pharmacological therapies in Parkinson's-related dementia

Non-drug-based approaches can provide an alternative to drug-treatments without causing adverse effects or side-effects (Ballard et al., 2009; Hindle et al., 2018; McCormick et al., 2017b; Morrin, Fang, Servand, Aarsland, & Rajkumar, 2018; Sindhi & Leroi, 2013); therefore, they may play an important role in people with PD and cognitive impairment. For motor and non-motor symptoms of PD, the suggested therapies include physiotherapy, speech and language therapy, occupational therapy, PD specialist nurse interventions (NICE, 2017), exercise, dance and multidisciplinary care interventions (Bloem, de Vries, & Ebersbach, 2015), mind-body exercises such as tai chi and yoga (Kwok, Choi, & Chan, 2016), exergames (where exercising is merged in the video games; Barry, Galna, & Rochester, 2014), gait training and virtual reality (Mirelman et al., 2011) as well as acupuncture, reflexology, education and music therapy (Ahn, Chen, Bredow, Cheung, & Yu, 2017). However, the level of motor impairment can act as a barrier for many people with PD and exercise therapy has the risk of causing injury.

Non-pharmacological approaches, that target behavioural, psychological, social and cognitive symptoms of neurodegenerative conditions without using medication, are often referred to as psychosocial interventions (Brodaty & Arasaratnam, 2012). The most beneficial interventions for behavioural and cognitive symptoms in PD are exercise and cognitive training (Bloem et al., 2015), and cognitive behavioural therapy (Yang et al., 2012), as well as cognitive or physical rehabilitation, education and brain stimulation techniques (Hindle, Petrelli, Clare, & Kalbe, 2013). Specifically, these interventions targeted neuropsychiatric symptoms (i.e. anxiety, depression, apathy), cognitive domains (i.e. attention, processing speed, memory, visuospatial abilities,

executive function, language, verbal fluency, and global cognition), and both motor and cognitive aspects (i.e. dual task performance) as well as activities of daily living and quality of life (Hindle et al., 2013; King et al., 2015; Yang et al., 2012). Importantly, when people with dementia were asked what they wished to attain from psychosocial interventions, people with dementia responded that the interventions should improve their well-being, health, confidence, social participation and human rights (Øksnebjerg et al., 2018). These domains need to be considered if the intervention is to be person-centred, otherwise people with dementia may not engage with the therapy. As some of the aforementioned interventions primarily target cognition, they will be discussed in detail in the next section.

2.1.6 Psychosocial interventions focused on cognitive impairment

Psychosocial interventions that focus on cognitive impairment and dementia are non-pharmacological interventions that aim to improve cognitive functioning either directly or indirectly (Bahar-Fuchs, Clare, & Woods, 2013). These interventions commonly include cognitive rehabilitation, cognitive training and cognitive stimulation (see Table 2.3) and can be delivered individually, in a dyad, in a group or online (Clare & Woods, 2004). Several studies have evaluated these specific types of interventions among people with PD (Costa et al., 2014; Farzana et al., 2015; Hindle et al., 2018); however, more rigorous studies are required to determine their efficacy. Indeed, Goldman and colleagues (2018b) emphasised that despite existing and on-going interventions that focus on cognitive impairment of people with PD, there remain several unanswered questions. For example, what cognitive tasks can be beneficial for people with PD, which specific cognitive domains are enhanced, whether positive changes in one cognitive domain affect other cognitive processes, and what neurobiological components determine the effectiveness of interventions targeting cognitive impairment, which should be the focus in future studies (Goldman et al., 2018b). The authors commented that large-scale, longitudinal, randomised controlled trials (RCT) with people at varying stages of PD should be conducted to study the impact of cognition-focussed interventions on prevention, delay and improvement of cognitive and functional domains (Goldman et al., 2018b). Hence, trialling specific interventions

that target cognitive impairment among people with PD-MCI, PDD and DLB is 'both timely and appropriate' (Mohlman, Chazin, & Georgescu, 2011).

Table 2.3 Comparisons between cognitive stimulation, cognitive training and cognitive rehabilitation (Bahar-Fuchs et al., 2013; Clare & Woods, 2004).

	Cognitive stimulation	**Cognitive training**	**Cognitive rehabilitation**
Aim	Enhance cognition & social function	Maintain & enhance cognitive functioning	Improve daily functioning and enhance well-being
Focus of intervention	A range of generic mentally stimulating activities & discussion	Guided practice on standardised tasks of cognitive memory, attention, or executive function	Set goals to identify functional needs (required to perform everyday tasks) & develop techniques to address these needs
Context	Cognitive tasks in a social setting	Structured tasks & environments	Real-world setting
Format	Individualised or group	Individualised or group	Individualised
Goals	Improvements in cognition and behavioural symptoms	Improved or maintained ability in specific cognitive domains	Performance & functioning in relation to collaboratively set goals
Evidence-base in dementia and in PD	Improves global cognition, quality of life, communication & social interaction in people with dementia No trials in PD	Improves executive functioning, working memory & processing speed in people with dementia and people with PD	Can improve self-rated competence, quality of life & memory capacity in people with dementia and people with PD
Evidence-based in Parkinson's-related dementia	One trial in PD-MCI (Farzana et al., 2015)	Two trials in PD-MCI (Costa et al., 2014; Reuter et al., 2012)	One pilot trial in PDD and DLB (Hindle et al., 2018)

Abbreviations: DLB – dementia with Lewy bodies; PD – Parkinson's disease; PDD – Parkinson's disease dementia; PD-MCI – Parkinson's disease with mild cognitive impairment.

2.1.6.1 Cognitive rehabilitation

Cognitive rehabilitation was first developed to address the effects of traumatic brain injury (Cicerone et al., 2000) and has gained popularity in dementia research over the last two decades (Bahar-Fuchs et al., 2013). The primary goal of *rehabilitation* is to help people find ways to manage their physical, mental and social functioning following an illness, trauma or injury in order to optimise their functioning (Clare & Woods, 2004). *Cognitive rehabilitation* aims to support people with cognitive decline in improving their daily functioning through goal-setting and implementation of a tailored plan to address the goals using evidence-based strategies (Bahar-Fuchs et al., 2013; Clare, 2017; Clare & Woods, 2004; Wilson, 1997). The intervention is highly individual and tailored according to each person's symptoms, impairments, needs, situational factors and preferences (Clare et al., 2010). Participants' goals may relate to everyday functioning, activities of daily living, personal care, language or social interaction. They are discussed and established with a trained therapist who works closely with every individual (Clare, 2017). The role of the therapist is crucial in order to: (1) define purposeful, feasible, reasonable, realistic and attainable goals, (2) assess the person's strengths and the resources required to achieve those goals, (3) aid the development of an action plan and provide strategies how to achieve the goals, and (4) act as a first point of contact and provide emotional and psychological support (Clare, 2017).

The term 'cognitive' can be ambiguous or confusing as cognitive rehabilitation focuses on improving everyday functioning and activities of daily living, rather than improving cognitive performance, but it is specifically targeted to people with cognitive impairment (Clare, 2017); thus, the term 'cognitive rehabilitation' is used to distinguish it from physical rehabilitation (Clare et al., 2010). Although traditionally, cognitive rehabilitation has been classified as one of the cognition-based interventions, more recently the focus has been on considering this intervention as targeting functional disability associated with cognitive impairment (Clare, 2017). To date, some evidence for effectiveness of cognitive rehabilitation has been found amongst people with PD (Alzahrani & Venneri, 2018), dementia (Clare et al., 2010; Kudlicka et al., 2017) and

PRD (Hindle et al., 2018); however, due to the limited number of studies, definite conclusions cannot be drawn.

2.1.6.2 Cognitive training

Cognitive training (also referred to as 'retraining', 'brain training' or 'remediation') appears to be one of the most widely researched cognition-based interventions in dementia (Bahar-Fuchs et al., 2013) and PD (Hindle et al., 2013). Cognitive training includes guided and repeated practice of selected standardised tasks addressing specific cognitive domains, such as memory, attention, executive function, language and speed of processing (Bahar-Fuchs et al., 2013; Clare, 2003; Clare, 2017; Clare & Woods, 2004; Goldman et al., 2018b; Walton et al., 2017). The intervention can be facilitated using either a pen-and-pencil method or a computer, and delivered in a group, individually or guided by a family member with support from a therapist (Bahar-Fuchs et al., 2013; Clare & Woods, 2004; Walton et al., 2017).

Cognitive training aims to enhance cognitive functioning through regular practice over time by using specific methods, for instance remembering, planning, focusing attention or organising information (Bahar-Fuchs et al., 2013; Clare & Woods, 2004; Goldman et al., 2018b). Positive results have been found for people with PD and normal cognition in terms of improving working memory, executive function and processing speed with cognitive training programs (Leung, Walton, Hallock, Lewis, Valenzuela, & Lampit, 2015). With the development of technology, studies have increasingly prioritised computer-based cognitive training, where exergames, cognitive games, and virtual reality have been incorporated (van de Weijer, Hommel, Bloem, Nonnekes, & de Vries, 2018). Researchers argue that methods involving computers may be more cost-effective, flexible, scalable and adaptable than non-computer based methods (Van de Weijer et al., 2018) and trials of their effectiveness should continue in the future. Despite the positive results of cognitive training for cognition and activities of daily living for people with dementia (Bahar-Fuchs et al., 2013; Huntley, Gould, Lou, Smith, & Howard, 2015) and people with PD (Leung et al., 2015), cognitive training is not supported by the NICE guidelines (NICE, 2018b) and there are no studies

supporting its use among people with PDD and DLB (Orgeta et al., under review). Lack of cognitive training studies in PDD and DLB could potentially be due to higher levels of disability, concerns over safety and well-being, and additional disease-related symptoms and needs among people with PRD.

2.1.6.3 Cognitive stimulation

Cognitive Stimulation Therapy (CST) was formed by systematically reviewing and analysing the literature to identify non-medicinal therapies for dementia (Spector, Davies, Woods, & Orrell, 2000; Woods, Spector, Jones, Orrell, & Davies, 2005). The authors identified and merged the most effective components of these therapies, including *Reality Orientation* (Taulbee & Folsom, 1966), *Reminiscence Therapy* (Butler, 1963), and *Cognitive Stimulation* (Breuil et al., 1994), to form a new, evidence-based CST programme (Spector, Orrell, Davies, & Woods, 2001). CST invites the person with dementia to join in a discussion about a range of topics, usually through group activities and interactions, which can include current affairs, food, childhood, the present day or word games (Bahar-Fuchs et al., 2013; Spector et al., 2001; 2003). The main focus of CST is to enhance cognitive and social functioning by doing socially and cognitively stimulating tasks, which in turn can lead to changes in cognition and, at times, in behaviour (Clare & Woods, 2004; Spector et al., 2000, 2003, 2010).

A multi-centre single-blind RCT of seven-week CST conducted at day centres and residential care homes with 201 people with dementia showed that cognition and quality of life had significantly improved in the treatment group compared to the control group, and positive trends in communication scores were seen in the CST group (Spector et al., 2003). Following the study, the authors were interested in the specific mechanisms of action and found that in the treatment arm, language (i.e. word-finding, comprehension and naming) had improved but memory and orientation remained unchanged, which could be due to the focus on opinions and stimulating language via categorisation (Spector et al., 2010). The qualitative findings support the efficacy of CST as participants felt more relaxed, confident and positive, finding it easier to communicate and feeling less lonely (Spector, Gardner, & Orrell, 2011). The

people with dementia, care partners and facilitators of the group therapy sessions also reported changes in cognition of people with dementia such as improved memory, concentration, alertness, attention as well as positive feelings and experiences of being in a supportive, non-judgmental and non-threatening group, which allowed people to express their personal views and share common difficulties (Spector et al., 2011). CST has also been found to be cost-effective compared to treatment as usual (Knapp et al., 2006), to improve quality of life (but not cognition) over six months, and improve cognition when combined with dementia medication (Orrell et al., 2014). The authors argued that cognitive decline did not reduce during an extended period of CST due to inevitability of dementia progression and they recommended that there may be a role for combining medicinal and non-medicinal treatments to improve outcomes for cognition (Orrell et al., 2014).

In 2012, a Cochrane review of CST for people with dementia was conducted consisting of 718 participants (407 people in the CST arm and 311 people in the control arm) in fifteen RCT's (Woods, Aguirre, Spector, & Orrell, 2012). The authors concluded that cognitive stimulation was beneficial in terms of cognition for people in mild-moderate dementia and the effects were maintained between one to three months after the therapy (Woods et al., 2012). Similarly, the quality of life and well-being of people with dementia had improved and the staff-rated level of communication was higher and social interactions were more frequent after participating in CST (Woods et al., 2012), suggesting that CST can bring both cognitive and social benefits to people with dementia (Spector et al., 2011). However, the authors noted that there was no difference in mood, behavioural symptoms and activities of daily living among people with dementia.

Given that CST can have positive effects on cognitive and social functioning, NICE guidelines recommend that structured group tasks such as CST should be provided to people with mild to moderate dementia, whereas cognitive training should not be offered to this group of people (NICE, 2018b).

Recently, an individualised home-based carer-delivered form of CST (iCST; Orgeta et al., 2015a; Orrell et al., 2017) was trialled in a pragmatic multi-centred single-blind RCT. The iCST trial recruited people with a diagnosis of any type of dementia (as determined by DSM-IV criteria) and was suitable for those people with dementia who could not attend the group-CST due to travel or personal needs (i.e. physical disability, hearing or vision impairment) and preferences (i.e. not willing or unable to take part in a group setting) (Orgeta et al., 2015a). As a result of participating in the intervention, the dyadic relationship and care partners' quality of life improved; however, changes in cognition, quality of life, neuropsychiatric symptoms and care partner mental health were not observed (Orgeta et al., 2015a; Orrell et al., 2017). Nevertheless, dyads felt that their communication had improved following the intervention (Orgeta et al., 2015a), and qualitative interviews with the dyads support these findings as dyads appreciated having the interactions with one another and spending time together doing mentally stimulating and engaging tasks that were pleasurable (Leung, Yates, Orgeta, Hamidi, & Orrell, 2017). This provides evidence that psychosocial interventions, such as CST and iCST, can be effective in improving dyadic outcomes.

Syntheses evaluating the efficacy of cognition-based interventions in Alzheimer's dementia found that cognitive stimulation was effective in improving cognitive scores, whereas the same did not apply for cognitive training (Huntley et al., 2015; McDermott et al., 2018). In contrast, the most endorsed cognitive intervention for PD without cognitive impairment is cognitive training (Goldman et al., 2018b; Hindle et al., 2013). However, despite the increasing evidence-base of enhancing cognition with non-pharmacological interventions in PD, relatively few trials have been conducted to date and many have methodological limitations, such as lack of power, no randomisation, exclusion of active control arm and short follow-up window (Bloem et al, 2015; Hindle et al., 2013). Given that only two pilot psychosocial studies have been undertaken in PRD, one of goal-oriented cognitive rehabilitation for people with PDD and DLB (Hindle et al., 2018) and one of cognitive training in PD-MCI (Costa et al., 2014), there is an urgent need to conduct rigorous, evidence-based, controlled non-pharmacological trials with people with PDD (Hindle et al., 2013) and DLB (Connors et al., 2018).

2.2 Overview of care provision in Parkinson's-related dementia

2.2.1 Terminology and definition of care provision

It has been well recognised that we as human beings need to be loved, nurtured and cared for by others (Kittay, Jennings, & Wasunna, 2005). Caring for and supporting those dear to us is seen as a natural part of what makes us human (Carers UK, 2014). In fact, caring has been considered so normal and universal that Rosalynn Carter, the former first lady who passionately advocated for carers, said:

> *"There are only four kinds of people in this world:*
> *those who have been caregivers, those who currently are caregivers, those who will be caregivers, and those who will need caregivers."*
>
> *- Rosalynn Carter*

Prior to defining the term 'carer' and describing what care provision entails, a discussion of the terminology is pertinent. Generally, the term '*carer*' is used in the United Kingdom, Australia and New Zealand and the term '*caregiver*' in the United States, Canada and elsewhere. Care provision is divided into two categories: *formal*, which is a paid profession, and *informal*, where care is provided by unpaid family members, usually spouses, children, siblings, relatives, friends or neighbours. The term 'carer' originates from the 1970s and 1980s feminist researchers who aimed to increase the visibility of the role and the experiences of care provision in the home setting (Fine & Glendinning, 2005; Molyneaux, Butchard, Simpson, & Murray, 2011a). It is also an extensively used term in health and social care and 'remains a gateway through which services are accessed' (Molyneaux et al., 2011a, p. 425). Despite the extensive research with carers in the last half-century, the term 'carer' has been critiqued by several academics.

Pilgrim (1999) encouraged discontinuation of the term 'carer' and suggested acknowledging the specific roles, context and relationships that people were in. He

further echoed that providing care does not necessarily equate with caring about the person (Pilgrim, 1999) despite the general understanding that the care provider does care about that person (Pearlin, Mullan, Semple, & Skaff, 1990). Furthermore, in Henderson's qualitative study (2001), a care recipient saw the notion of referring to one's partner as 'a carer' unacceptable because being in both roles simultaneously was not feasible and could significantly impact a couple's relationship. Molyneaux and colleagues (2011a) suggested re-evaluating the term 'carer' and potentially dropping the term 'carer' as it does not have 'carers' and 'care recipients' at its core. Even though it would be complex to change the use of this term worldwide, the authors recommended that clinicians and researchers should clarify what they mean when referring to 'carers' and 'caregiving' (Molyneaux et al., 2011a).

Several other terms, such as care provider, caretaker and care partner, have been used to refer to informal, lay, unpaid or untrained carers (Smith, 2001). These terms are widely used in research due to their coherence, clarity and popularity; however, they do not take into account the care dyad and the care relationship – the bond between the person receiving care and the person providing it (Bennett, Wang, Moore, & Nagle, 2017; Eilers, 2013; Kittay et al., 2005). Kittay and colleagues (2005, p. 444) said that: "All caregiving involves a direct, intimate relationship between two or more people", which takes place in a psychosocial context. This notion is corroborated by Eilers (2013) who described that the care partnership is built on pillars of trust, equality and shared experience. The caring experience is global, varying from deeply personal experiences and emotional involvement to doing a purposeful activity (Kittay et al., 2005). In order to acknowledge the two-directional partnership and a caring relationship, the term '*care partner*'[1] will be used throughout this thesis.

The care partner is an individual, usually a spouse or an adult child, who has taken on the responsibility to help, support and assist a family member who cannot take care of themselves, and to assure they are safe and well (Ham, 1999; Pearlin et al., 1990). Care

[1] This thesis will specifically focus on those care partners who are spouses or long-term partners, collectively referred to as 'care partners' or 'life partners' of people with Parkinson's-related dementia.

provision helps the person with the condition to reach the highest functioning possible in their daily life (Ham, 1999). Often, the care partner supports with personal, psychological and medical care, assisting with mental and physical exercising, maintaining good nutrition, arranging living conditions and helping with housework (Brodaty & Donkin, 2009; Ham, 1999; Hand, Oates, Gray, & Walker, 2018; McLaughlin et al., 2010). Care partners also coordinate, plan and manage care and look for various interventions and treatments that could potentially alleviate the symptoms of the care recipients (Brodaty & Donkin, 2009; Ham, 1999). Notably, in addition to providing care, a proportion of care partners may be in part-time or full-time employment (Ostwald, 1997), which raises complex issues around managing their work and care commitments and may diminish their time and energy to provide care. In addition, care partners may also be older adults themselves and have physical and mental health issues which may limit their capabilities to provide care (Hand et al., 2018). As a consequence, care partners, particularly within dementia, may have increased negative feelings, depression, diminished well-being, and neglect their own health (Pinquart & Sörensen, 2003a). Thus, they become 'the invisible or hidden patients' (Brodaty & Donkin, 2009; Fengler & Goodrich, 1979; Ostwald, 1997).

A difference exists between caring and caregiving. Namely, caring is the affective component of "one's commitment to the welfare of another", whereas caregiving is "the behavioural expression of this commitment" (Pearlin et al., 1990, p. 583). Likewise, caring has been described as the interplay between emotion and action involving endearing feelings such as love as well as activities involving labour (Finch & Groves, 1983; Hennings, Froggatt, & Payne, 2013). In fact, providing care has even been named as the 'unexpected career' due to the sudden onset of this role (Pearlin & Aneshensel, 1994). The shift into taking on care responsibilities may either be gradual or sudden, depending on the level of independence and functionality of the person requiring care. However, it is common that many people do not plan or prepare for this role as it is accidental, unforeseen, unpredictable and unintentional; therefore, it can be difficult to accept or ease into the role (Pearlin & Aneshensel, 1994).

Entering the 'care provision' territory can be frightening as it is new, unknown and unfamiliar, which requires endurance, persistence, stamina, tolerability, high adaptability, flexibility, resilience and patience from care partners. Frequently, caring may take place in the context of a marital relationship or long-term partnership, but this can be protective because spouses who provide care to their partners see it as a natural extension of their love and a normal course in their relationship (Gaugler, Kane, & Kane, 2002; Gillies, 2011; Lawn & McMahon, 2014; Martin, 2016; Molyneaux, Butchard, Simpson, & Murray, 2011b). Due to the progressive nature of neurodegenerative conditions, the presenting symptoms may be so subtle that care partners may not notice a visible change in their responsibilities, even though they may have started to help and support the care recipients. Thus, in early stages of the disease, care partners may not identify themselves as such and may even dislike being called a 'carer' (Gaugler et al, 2002; Leroi, 2017; Molyneaux et al., 2011b; Pearlin & Aneshensel, 1994), rather, they prefer to be acknowledged as a 'spouse', 'partner' or 'support person' (Leroi, 2017). Therefore, it is important to acknowledge the relationship between the person receiving care and the person providing it.

2.2.2 Facts and figures of care partners

Around the world, one person in ten is a care partner [Office for National Statistics, (ONS), Census 2011]. In the UK, there are currently 6.5 million people who provide care and each day 6,000 people in addition take on the caring role (Carers UK, 2014). Of all the care partners in Great Britain, approximately 11% provide care to someone with dementia in a home setting (Carers Trust, 2015). Financially, the contribution that carers make exceeds £132 billion per annum, which surpasses the annual budget of the National Health Service (NHS) in England (Carers UK, 2018), showing that the help and support that care partners provide is invaluable and has cost-saving implications for the health and social care system.

In the UK, about 58% of care partners are female and women are more likely to accept a care partner role than men (ONS, 2011). Women may frequently become care partners of older people (Braithwaite, 1992), people with PD (Hand et al., 2018),

dementia (Brodaty & Donkin, 2009) and DLB (Galvin et al., 2010). Braithwaite (1992, p. 22) argues that "women are particularly at risk, not only because they are more likely to become caregivers, but because they are being denied the opportunity to be something other than a caregiver". Furthermore, female care partners may neglect their own needs and not receive opportunities to develop outside of their caring role and participate in activities, as men would (Braithwaite, 1992). Female spousal care partners of people with dementia have experienced higher strain and burden (Brodaty & Donkin, 2009; Hooker, Manoogian-O'Dell, Monahan, Frazier, & Shifren, 2000) than male care partners, although gender differences were not found in care partners of people with PD (Hooker et al., 2000). This suggests that changes in cognition play a significant role in determining outcomes in male and female care partners. In terms of age, even young spousal care partners of people with PD (i.e. aged between 40 and 55) were finding their caring role difficult and burdensome compared to those who were older (i.e. above 70 years) as they were working and raising children concurrently to their caregiving responsibilities (Carter, Lyons, Stewart, Archbold, & Scobee, 2010). In short, these findings underscore that providing care to someone with PD and related cognitive impairment may be particularly complex and has a direct impact on care partners' health, well-being and life.

2.2.3 Policies in support of care partners

In 2018, two important reports were published in the UK: the State of Caring 2018 (Carers UK, 2018), which summarises the thoughts and needs of care partners, and the Carers Action Plan for 2018-2020, which describes the actions that the government plans to take in the near future. Specifically, in the State of Caring 2018 report, care partners wished that carers would be routinely identified and supported; would receive appropriate information, training and equipment; and would get proper breaks (Carers UK, 2018). Similarly, these priorities overlap in the government's action plan: (1) health and social care services should be responsive and flexible to care partners and ensure they are included, valued and supported, (2) care partners should be recognised in the wider community and society, and (3) research should continue strengthening the evidence-base and seeking effective solutions to improve outcomes

for care partners (Department of Health and Social Care, 2018). Moreover, the government recognises that care partners are important in the health and social care system but despite this many care providers wished to be more actively involved in the care of their family members (Carers UK, 2018), which should be continuously encouraged by healthcare professionals.

The Care Act 2014, which is the most recent act acknowledging the needs of care partners, has elucidated that local councils should advance the well-being of those who provide care, offer financial advice and provide a carer assessment to evaluate their needs. If a care partner is trained, supported and informed, they are the most significant leader in the healthcare team as they are, in most cases, involved for the entire duration of the illness (Ham, 1999). In line with the government's commitment to increase support for care partners, it is imperative that the care partners, healthcare services and the government work together to provide the help and support that care partners need.

In 2017, the World Health Organisation (WHO) published a global action plan for the years 2017-2025. One of the main action points focuses on supporting care partners of people with dementia with the following steps (WHO, 2017):

(1) Provide accessible and evidence-based information, training programmes and respite services to care partners to improve knowledge and caregiving skills;
(2) Train healthcare and social care staff to identify and reduce care partner stress and burn-out;
(3) Strengthen care partner protection, including social and disability benefits, policies and legislation against discrimination;
(4) Involve care partners in the planning of care.

The WHO aspires that by 2025, 75% of countries would provide support and training programmes for care partners of people with dementia. Likewise, the Organisation for Economic Co-operation and Development (OECD; Colombo, Llena-Nozal, Mercier, & Tjadens, 2011) has prompted augmentation of policies across OECD countries to

support care partners. OECD also contended that care partners will remain in their role longer if they feel valued. However, studies showed that many care partners of people with PD had not received appropriate information, advice and support, and were not involved in the care plan of their family members (McLaughlin et al., 2010; Theed, Eccles, & Simpson, 2017). This has implications for the future care of care partners and it has been recognised for PD care partners as well. Specifically, the Parkinson's UK policy statement (2017c) affirmed the importance of focusing on the care partners, providing targeted help and support (including respite care and annual health checks), assessing their health needs and finding suitable treatment solutions. Continuous work by government, health and social care professionals and voluntary sector organisations (e.g. Parkinson's UK) can lead to better outcomes and support for care partners, who as a result may be healthier and able to continue providing care to their partners for longer.

2.2.4 Care provision in Parkinson's-related dementia

A growing body of research spanning several decades has drawn attention to the impact that PD has on care partners (Greenwell, Gray, van Wersch, van Schaik, & Walker, 2015; Mosley, Moodie, & Dissanayaka, 2017). It is well established that the progressive and complex nature of the motor, psychiatric and cognitive symptoms of Parkinson's (Chaudhuri et al., 2006) can reduce one's ability to carry out everyday activities and take care of oneself, thus increasing the need of a care partner. Care partners have a substantial role to play in the lives of people with PD and PRD as they support and assist with activities of daily living, personal care, medication, feeding, housework, attending specialists' appointments, maintenance of the person's quality of life and independence (Galvin et al., 2010; Hand et al., 2018; Hiseman & Fackrell, 2017; Tan, Williams, & Morris, 2012). The involvement of a care partner in the care of their family member is advantageous because they have a unique perspective on their partner's condition and thus, can provide a more precise and detailed description of their partner's symptoms (Hiseman & Fackrell, 2017). Notably, including care partners is so imperative that Brodaty and Donkin (2009) contended that without the help of care partners, the quality of life of people with neurodegenerative conditions would

drop so much that it would increase admissions to institutional care. This comes at the cost of care partners' own quality of life (Brodaty & Donkin, 2009) and raises an important question about how to maintain the well-being of both partners when facing a neurodegenerative condition.

Providing care to a person with Parkinson's can be emotionally draining, physically challenging and mentally exhausting for care partners (Aarsland, Larsen, Karlsen, Lim, & Tandberg, 1999a; Roland, Jenkins, & Johnson, 2010; Tan et al., 2012). The impact of PD on care partners is multifaceted, including social, financial, physical, emotional, mental and cognitive aspects. Socially, care partners of people with PD may not be able to go out as much as before, struggle to get away on holidays and have fewer social interactions with their friends, family and neighbours (Galvin et al., 2010; O'Reilly, Finnan, Allwright, Smith, & Ben-Shlomo, 1996; Schrag, Hovris, Morley, Quinn, & Jahanshahi, 2006; Thommessen et al., 2002). In addition, due to care provision many care partners may be unable to do their usual daily tasks, activities and hobbies, and may receive insufficient social support from friends and family. Having hobbies, being socially engaged and active and receiving social support are important because they could protect against worsening of health and well-being (Berger et al., 2017; Chappell & Reid, 2002; Greenwell et al., 2015). Physically, care partners may experience deterioration in health (O'Reilly et al., 1996), health-related quality of life (Lawson et al., 2017; Leroi, McDonald, Pantula, & Harbishettar, 2012a; Martinez-Martin et al., 2008) and greater fatigue (Aarsland et al., 1999a).

In terms of mental-emotional aspects, care partners may encounter negative feelings, such as frustration, sadness, anger, resentment, guilt, worry (Aarsland et al., 1999a; Tan et al., 2012), and feel overwhelmed, stressed, strained and burdened (Carter, Stewart, Lyons, & Archbold, 2008; Galvin et al., 2010; Leiknes, Lien, & Severinsson, 2015; Lökk, 2008; Martinez-Martin et al., 2005, 2008, 2015; Miller, Berrios, & Politynska, 1996; Mosley et al., 2017; Whetten-Goldstein, Sloan, Kulas, Cutson, & Schenkman, 1997). Care provision may significantly increase anxiety and depression (Aarsland et al., 1999a; Martinez-Martin et al., 2008; Schrag et al., 2006) and lower care partners' mental health (Peters, Fitzpatrick, Doll, Playford, & Jenkinson, 2011). As

a consequence, PD care partners' life satisfaction may reduce (Aarsland et al., 1999a). Furthermore, in non-PD care partners, the rates of mortality (Schulz & Beach, 1999), cognitive impairment (Mallya & Fiocco, 2018) and relationship dissatisfaction (Steadman, Tremont, & Duncan Davis, 2007) may increase. All of these factors can be escalated with the progression of cognitive impairment in PD (Roland & Chappell, 2017), which suggests focusing on the care partners of people with PRD is crucial.

The profile of care partners of people with PD has recently been described. Commonly, a care partner of a person with PD is a female spouse, aged around 70 years, living with her partner, having provided care for an average of 5 years and currently providing up to 16 hours of care per day (Cifu et al., 2006; Hand et al., 2018; Lökk, 2008; Martinez-Martin et al., 2015; Peters et al., 2011). Although these descriptions are comparable to those providing care to someone with dementia, the care provision hours in PD are notably higher than in dementia (i.e. 6-9 hours per day) (Brodaty & Donkin, 2009). The typical tasks that care partners helped people with PD with were assisting with household chores (i.e. cleaning, washing cooking), being there as a partner and friend (i.e. listening, providing support), personal care (i.e. bathing, dressing), and feeding and helping at night (Hand et al., 2018). A recent qualitative meta-synthesis summarised the experiences of PD care partners into four interrelated themes describing (1) the need to carry on as usual, (2) the importance of support in facilitating coping, (3) the difficult balancing act between caregiving and caregiver needs, and (4) conflicts in seeking information and knowledge (Theed et al., 2017). Thus, care provision within PD has been considered unique and complex in comparison to other neurodegenerative conditions. However, little is known about the profile of care partners of people with PDD and DLB. Therefore, one of the aims in this thesis was to describe the profile of PRD care partners (see Study 1, Chapter 5).

Studies have evaluated what aspects of PD (in the absence of cognitive impairment) have the highest impact on care partners. Findings suggest that both motor and non-motor symptoms of PD affect care partners' well-being, quality of life and burden but non-motor domains, particularly psychiatric manifestations such as apathy, psychosis, depression and cognitive impairment, tend to have a stronger effect (Aarsland et al.,

1999b, 2007; Carter et al, 2008; Greenwell et al., 2015; Leiknes et al., 2015; Martinez-Martin et al., 2015; Mosley et al., 2017; Schrag et al., 2006). Similarly, the notion that caring for someone with mental illness is emotionally harder, more complex and taxing, as opposed to caring for someone with a physical illness, has been depicted before, which may be due to the changeable, unstable and erratic symptom presentation in mental health conditions, which disrupts 'the coherence of everyday life' (Karp & Tanarugsachock, 2000 p.7). This is in line with literature on care partners of people with dementia (Brodaty & Donkin, 2009; Zarit, Todd, & Zarit, 1986), PD (Greenwell et al., 2015; Lawson et al., 2018) and DLB (Galvin et al., 2010), confirming once again the complexity of non-motor symptoms in PRD. Considering that research with care partners of people with PD has mostly examined the impact of PD on burden and stress, physical and mental health, well-being and quality of life, these constructs were chosen as outcomes of interest in the studies included in this thesis and will be described in more detail in the following sections.

2.2.4.1 Burden in care partners

One of the most researched constructs in care partner research is 'caregiver burden' (van der Lee et al., 2014). Several different definitions have been proposed but two interwoven descriptions from the 1980s are used concurrently to this day. George and Gwyther (1986, p. 253) define burden as "the physical, psychological or emotional, social, and financial problems that can be experienced by family members caring for impaired older adults". The same year, Zarit *et al.* (1986, p. 261) proposed a very similar explanation adding that burden is "the extent to which caregivers perceive their emotional or physical health, social life, and financial status as suffering as a result of caring for their relative". Even though both explanations encompass the multifaceted impact on care partners, the definitions of burden are still diverse, incoherent and vague in many research studies making measuring 'burden' ambiguous (Bastawrous, 2013; Braithwaite, 1992). The authors recommend that burden should be defined clearly, researched using mixed methods (i.e. both quantitatively and qualitatively) and evaluated as specific dimensions of burden (Bastawrous, 2013; Braithwaite, 1992). The factors of burden are explored in Study 2 in this thesis.

In line with earlier discussions about the accuracy of the term 'burden', the term has been critiqued and is becoming less favoured, which could be due to several reasons. On one hand, the person receiving care may feel as if they are a burden, and on the other hand, the person providing care may not necessarily experience it as a burden but rather as an extension of marital commitment and moral responsibility (Carl, 2017; Kilgariff & Grant, 2016). Care partners have also preferred the use of term 'strain' as it describes their experience of caring more precisely (Abendroth, Lutz, & Young, 2012). Thus, it is suggested that we endorse the relationship of the dyad (Pilgrim, 1999), as mentioned earlier. Notwithstanding the decreased acceptance of the term 'burden', it remains a popular term in the literature and therefore will be used throughout this thesis for consistency with earlier research.

Burden has been characterised as a highly subjective experience which varies between individuals (Poulshock & Deimling, 1984). However, an 'objective' burden has also been recognised, which includes physical care provision and helping with activities of daily living (i.e. measured in hours that care was provided for) (Bastawrous, 2013; Montgomery, Gonyea, & Hooyman, 1985). In contrast, the subjective burden is the psychological impact and emotional reaction that care provision has on care partners (i.e. assessed with self-rated well-being, burden and strain scales) (Bastawrous, 2013; Montgomery et al., 1985). Given that objective and subjective burden are frequently assessed within one scale, it remains unclear whether they are separate domains or predictive of one another (i.e. hours of care can contribute to negative feelings in the care partner), which can prevent interpreting the burden construct clearly (Bastawrous, 2013).

In PD, several different terms exist to refer to burden, for instance *strain* (Kelly et al., 2012; Lökk, 2008; Martinez-Martin et al., 2005; Nygaard, 1988), *stress* (McRae, Sherry, & Roper, 1999) and *distress* (Lau & Au, 2011; Miller et al., 1996). Despite the fact that these terms have been used instead of burden or in conjunction with burden (Leiknes et al., 2015), recent studies have determined that these constructs are independent from burden and are evaluated as separate constructs (Cifu et al., 2006; Leiknes et al., 2015; Mosley et al., 2017; Santos-Garcia & de la Fuente-Fernandez, 2015). As

Parkinson's progresses, the cognitive impairment advances leading to higher *strain* (Carter et al., 2008), *burden* (Cifu et al., 2006; Jones et al., 2017; Leroi et al., 2012a; Martinez-Martin et al., 2015; Szeto et al., 2016) and *stress* (Aarsland et al., 2007) in care partners. The main contributors to care partner burden and stress in people with PDD were the person's neuropsychiatric symptoms (i.e. apathy, depression, psychotic symptoms) (Aarsland et al., 2007; Martinez-Martin et al., 2015) and cognitive decline (Cifu et al., 2006; Jones et al., 2017; Leroi et al., 2012a). This describes the unique nature of the neuropsychiatric profile in PDD (Aarsland et al., 2007) and DLB (Galvin et al., 2010), compared to other types of dementia, and underscores that PD-MCI, PDD and DLB should be evaluated jointly due to similarities in clinical symptom presentations and their impact on care partners.

2.2.4.2 Physical and mental health in care partners

The State of Caring 2018 survey in the UK (Carers UK, 2018) found that 72% of care partners experienced worsening of their mental health and 61% in their physical health due to their caring role. Furthermore, over half of care partners anticipated that both physical and mental health would continue to deteriorate over the coming years, and a third of participants predicted that a decline in their mental and physical health would prevent them from being able to provide care to the care recipients in the future (Carers UK, 2018). Providing care to someone with a neurodegenerative condition can have a significant impact on the care partner's health (Pinquart & Sörensen, 2007). Several intrapersonal and interpersonal aspects, for example own depression, higher age, lower socioeconomic status, lack of social support and care recipients' behaviour, were associated with poorer physical health in care partners (Pinquart & Sörensen, 2007). Among PD care partners, over a third experienced a deterioration of their health due to care provision (Schrag et al., 2006). Lack of sleep, fatigue, high blood pressure, muscle strain, headaches and gastrointestinal problems were also common in this group (Lökk, 2008). These health issues had developed as a result of providing care.

Many studies have identified that care provision within PD can worsen mental health in care partners. In fact, the mental health of those who care for people with PD is poorer (Aarsland et al., 1999a; Peters et al., 2011) and distress greater (Martinez-Martin et al., 2008) compared to the general population. A variety of scales have been employed to measure mental health among care partners of people with PD, including global mental health, depression, anxiety and stress scales (Greenwell et al., 2015). Up to half of care partners of people with PD can experience clinically significant anxiety and depression (Mosley et al., 2017). Poor mental health in care partners is directly linked to duration of care provision in years and proportion of hours devoted to caring each day (Peters et al., 2011). Moreover, lower levels of mental health are also predicted by partners' motor, psychiatric and cognitive symptoms, although drawing definite conclusions about what predicts mental health remains difficult due to the variability of the measures, inconsistent findings and lack of evidence (Greenwell et al., 2015). Importantly, despite the care partners' own health needs, they felt they had to stay healthy as long as possible to be able to care for and support their partners (Berger et al., 2017; Tan et al., 2012). This presents major physical, financial, emotional, mental and social challenges for care partners to continue in their role whilst taking care of themselves.

2.2.4.3 Quality of life in care partners

Providing care to a person with PD can have a direct effect on care partners' well-being and quality of life. In the literature, quality of life has been synonymously used with other terms such as health, health status, perceived health, functional status, and health-related quality of life although these terms are independent of one another (Martinez-Martin, 2017). The concepts of quality of life are wide incorporating economic, environmental, cultural, social, spiritual and personal aspects (Martinez-Martin, 2017; WHO, 1997), whereas health-related quality of life specifically focuses on individual's physical, mental and social aspects and the perceptions of their global health (Martinez-Martin, 2017; WHO, 1997). Health-related quality of life has been found to be lower among care partners of people with PD compared to general population (Martinez-Martin et al., 2008) and decreases with the emergence and

development of cognitive impairment in PD (Lawson et al., 2017; Leroi et al., 2012a; Szeto et al., 2016).

Quality of life is associated with several factors. Lower quality of life in care partners was predicted by the care recipients' disease-related factors (i.e. motor, cognitive and neuropsychiatric symptom severity, poorer quality of life, higher need for care, greater dependency in activities of daily living), personal aspects (i.e. higher age, depression) and care-related variables (i.e. longer duration of care provision in years and hours per day) (Greenwell et al., 2015; Hand et al., 2018; Lawson et al., 2017; Morley et al., 2012). Well-being of care partners is important because lower strain and 'caregiving load' reduces the risk of institutionalising persons with PD (Abendroth et al., 2012), which has long-term implications for the future.

2.2.4.4 Comparison of care partners' outcomes in neurodegenerative conditions

Comparative studies between care partners of different neurodegenerative conditions have shown important distinctions. In one study, burden in care partners was higher in PDD compared to AD, with neuropsychiatric disturbances fundamentally contributing to burden in care partners of people with PDD (Shin, Youn, Kim, Lee, & Cho, 2012). Another study supported these findings and added that care partners of people with PDD experienced more depression, lower satisfaction with life and needed more help and assistance compared to care partners of people with PD and AD (Roland & Chappell, 2017). Similarly, care partners of people with DLB had higher burden (Svendsboe et al., 2016) and distress (Bjoerke-Bertjeussen, Ehrt, Rongve, Ballard, & Aarsland, 2012; Ricci et al., 2009) compared to care partners of people with AD and frontotemporal lobar degeneration (Liu et al., 2018) due to more prominent neuropsychiatric symptoms in DLB. Care partners of both people with PDD and DLB also experienced higher levels of stress compared to AD and vascular dementia (Lee, McKeith, Mosimann, Ghosh-Nodyal, & Thomas, 2013). The researchers advised that more help and support should be provided for care partners of people with DLB, which is also applicable in PDD.

2.2.5 Theoretical framework of care provision

To understand the impact of PD factors on care partners and how they affect care partner well-being and the dyadic relationship, a theoretical framework is required. Such a framework also helps to understand the connections between the variables and to determine the direction of predictors. In the context of dementia, a number of multi-component models have been developed evaluating the factors contributing to caregiving-related stressors (van der Lee, Bakker, Duivenvoorden, & Dröes, 2014). The most common care partner stress models in dementia (van der Lee et al., 2014) are:

(1) The Transactional Model of Stress and Coping (Lazarus & Folkman, 1984);
(2) The Two-Dimensional Model of Psychosocial Morbidity (Poulshock & Deimling, 1984);
(3) The Stress Process and Coping Model (Haley, Levine, Brown, & Bartolucci, 1987);
(4) The Stress Process Model (Pearlin et al., 1990).

These four preceding models take into account the characteristics of each member of the dyad as well as the care recipient's disease symptomatology and care partner's reactions and outcomes. In PD, the **Stress Process Model** (Pearlin et al., 1990) and the **PD-specific Stress-Appraisal Model** (Goldsworthy & Knowles, 2008) are most applied by researchers. The Stress-Appraisal model (Goldsworthy & Knowles, 2008) has been built on previous similar models (Chappell & Reid, 2002; Lawton, Kleban, Moss, Rovine & Glicksman, 1989; Lawton, Moss, Kleban, Glicksman, & Rovine, 1991; Pearlin et al., 1990; Yates, Tennstedt, & Chang, 1999) and has since been developed further following a systematic review which evaluated burden, mental health and quality of life among care partners of people with PD (Greenwell et al., 2015). The proposed adaptations by Greenwell and colleagues (2015) are depicted in Figure 2.2.

The adapted Stress-Appraisal model by Greenwell et al. (2015), derived from the Stress-Appraisal model by Goldsworthy and Knowles (2008), was chosen as the theoretical framework for this thesis. This model captures care partners' experiences

of care provision in PD over and above other models (see Figure 2.2) and consists of four main domains (Goldsworthy & Knowles, 2008; Greenwell et al., 2015):

(1) **Stressors**: care partner well-being is affected by the person with PD factors (= primary stressors), such as neuropsychiatric and cognitive symptoms, their quality of life, their ability to perform activities of daily living and functional dependency (but not motor symptoms), which decreases physical health and increases depression in the care partner (= secondary stressors),

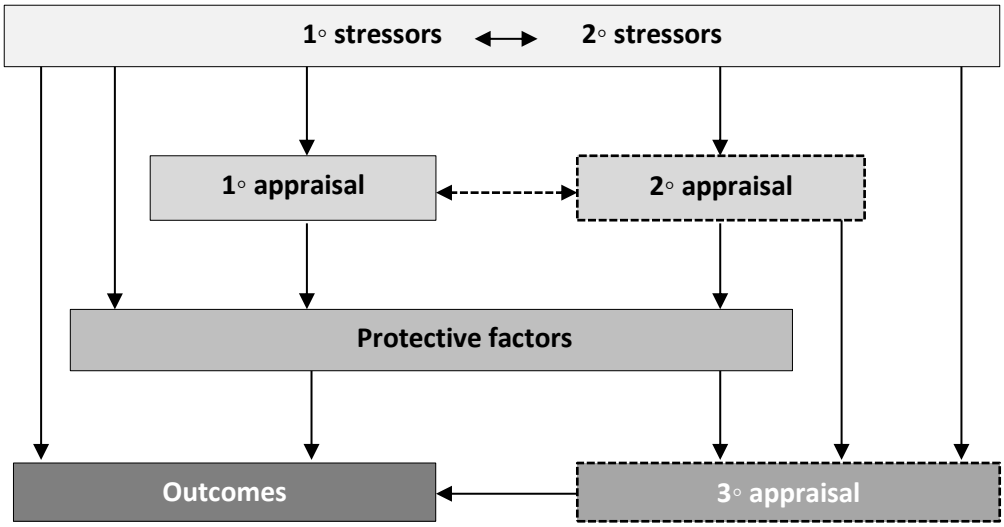
(2) **Stress appraisals**: how care partners experience the disease can influence whether they make 'primary appraisals' (i.e. seeing the disease as threatening and thus care partner becomes more involved in care provision by providing more hours of care) or 'secondary appraisals' (i.e. increase of burden and potentially developing coping strategies). Greenwell et al. (2015) proposed that 'tertiary appraisals', which are affected by primary and secondary stressors, 'secondary appraisal' and 'protective factors', also have a role in determining perceived burden and perceived uplifts by care partner, although burden was seen as a secondary appraisal in Goldsworthy and Knowles' (2008) model.

(3) **Protective factors (or mediators)**: an important predictor of burden is perceived social support, which can promote well-being or protect from negative consequences of stress. In Goldsworthy and Knowles' (2008) model, quality of dyadic relationship, frequency of breaks, formal service hours as well as care partner self-esteem were important mediators in the process of care partner stress appraisal. Greenwell *et al.* (2015) proposed that other predictors may include care partner personality traits, sense of coherence and self-efficacy, which require further investigation (Greenwell et al., 2015).

(4) **Outcomes**: the impact of primary and secondary stressors; primary, secondary and tertiary appraisals, and protective factors have a direct or indirect impact on care partner outcomes, such as determining their quality of life and depression.

The Stress-Appraisal model is useful in understanding the experiences of care partners in the context of PD and could be applied to PRD as well.

Figure 2.2 The Stress-Appraisal model adapted from Greenwell et al. (2015).



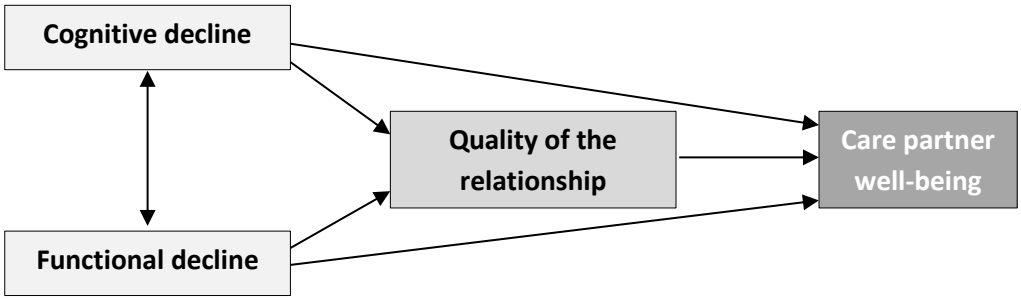
Legend: The dash line needs further examination. The dash boxes depict alterations to Goldsworthy & Knowles (2008) model by Greenwell et al. (2015).

Although the Stress-Appraisal model is comprehensive, it does not incorporate the dyadic relationship as an important factor in the context of caregiving relationship. Townsend and Franks (1995) proposed the **Binding Ties Theory**, which was designed to describe the quality of the relationship between adult children and their parents with cognitive impairment (see Figure 2.3). The authors considered the quality of the dyadic relationship to be crucial in care provision and an important determinant in the caregiving experience (Townsend & Franks, 1995). The model describes the associations between cognitive and functional impairment, closeness (positive), conflict (negative) and care partner well-being through measures of subjective caregiving stress, subjective caregiving effectiveness and depression. The findings suggest that negative ties were more predictive of care partner well-being than positive ties (Townsend & Franks, 1995). Furthermore, the pathway of 'cognitive decline → relationship quality → care partner well-being' was stronger than the 'functional decline→ relationship quality → care partner well-being' pathway and advancing cognitive impairment led to less closeness and more conflict in the dyadic

relationship (Townsend & Franks, 1995). This highlights that studies should evaluate both positive and negative interactions in the context of caregiving relationships.

In regards to the intimate dyadic relationship in PRD, the Townsend and Franks (1995) model could be incorporated in the Stress-Appraisal model (Goldsworthy & Knowles, 2008; Greenwell et al., 2015) by considering 'cognitive and functional decline' as primary stressors, 'quality of the relationship' as a protective factor and 'care partner well-being' as an outcome. Thus, this thesis will be based on the Stress-Appraisal model and include dyadic relationship quality as the mediator/protective factor in the model.

Figure 2.3 The Binding Ties Theory (Townsend & Franks, 1995).



2.2.6 Psychosocial interventions for care partners

In light of the effects that care provision can have on care partners of people with PRD, researchers have recognised the importance of psychosocial interventions specifically targeted to care partners. Since there are limited trials of psychosocial interventions for care partners of people with PD-MCI, PDD and DLB, this section will provide evidence from dementia and PD-research.

In dementia, the majority of interventions for care partners are individual or dyadic but also include telephone-based interventions (Brodaty & Arasaratnam, 2012). The most common psychological interventions for care partners of people with dementia are psychosocial, psychoeducational, technological, psychological (i.e. talking and discussing, cognitive behavioural therapy) and occupational therapies, as well as

support groups and multicomponent interventions (Gilhooly et al., 2016). Gilhooly and colleagues (2016) summarised that out of these, the most beneficial interventions for dyads of people with dementia and their care partners were psychosocial and psychoeducational therapies but support groups and multicomponent interventions have also been found effective (Gilhooly et al., 2016). Recently updated NICE guidelines, which summarise how to support people with dementia and care partners (2018b), also recommend psychoeducation and skills training interventions for care partners to educate about dementia, build care partners' skills, provide appropriate training in terms of care provision and disease symptoms, and give advice about self-care and well-being as well as help to plan for the future, which are also applicable to care partners of people with PRD.

Similarly to Gilhooly *et al.*'s findings (2016) and NICE guidelines (2018b), other systematic reviews have confirmed these findings and have supported the use of psychoeducational and multi-component interventions for care partners (Parker, Mills, & Abbey, 2008), social interventions with or without cognitive components (Cooke, McNally, Mulligan, Harrison, & Newman, 2001), and combining educational elements (e.g. increase care partners' knowledge of dementia and caregiving) with therapeutic aspects (e.g. cognitive behavioural therapy) (Dickinson et al., 2017; Sörensen, Pinquart, & Duberstein, 2002). A systematic review and meta-analysis of educational interventions for care partners of people with dementia endorsed educational programmes in terms of reducing burden and level of depression of care partners (Jensen, Agbata, Canavan, & McCarthy, 2015). Furthermore, online peer support groups (Hopwood et al., 2018; McKechnie, Barker, & Stott, 2014a; O'Connor, Arizmendi, & Kaszniak, 2014) and internet-based interventions (Boots, de Vugt, van Knippenberg, Kempen, & Verhey, 2014) have also been found advantageous for care partners, and have even improved relationship quality with the person with dementia (McKechnie et al., 2014a). As with interventions for people with dementia, many of these interventions for care partners can be delivered individually, in a group, online or as a dyad with the person with dementia; however, the evidence is mixed as to what delivery method is most efficacious for care partners.

The components of the aforementioned psychosocial interventions relate to various aspects of the adapted Stress-Appraisal model (Greenwell et al., 2015) as care partner involvement in the intervention describes *primary appraisals*, coping strategies that they have obtained refer to *secondary appraisals*, the relationship quality of the dyad and self-efficacy of the care partner relates to *protective factors* and finally the impact of primary and secondary appraisals and protective factors leads to *outcomes* such as reduced or heightened burden, quality of life or depression in care partners. It appears that both educational and therapeutic interventions aim to strengthen care partners' self-efficacy (i.e. *primary appraisal or protective factor*) but can also target *tertiary appraisals* and *outcomes* as one trial (Jensen et al., 2015) demonstrated that educational interventions reduced burden and level of depression among care partners. The internet-based support groups and interventions helped to increase relationship quality which acts as a *protective factor* according to the **adapted Stress-Appraisal model** (Greenwell et al., 2015). Dyadic cognition-specific interventions such as iCST could also have a *protective factor* as they can support dyadic relationship quality and could ultimately have a beneficial effect on care partners' outcomes.

The effectiveness of psychosocial interventions can be increased in several ways. Weinbrecht, Rieckmann and Renneberg (2016) advised tailoring the intervention and delivering face-to-face skills training. Parker and colleagues (2008) provided several specific recommendations to enhance interventions: (a) include both members of the dyad, (b) reinforce participation in educational interventions for care partners, (c) provide individualised programs, (d) support care partners regularly with appropriate information about their role as a care partner and inform them about available services, and (e) target behavioural symptoms in care recipients. Other researchers have suggested employing multi-component interventions and a technological aspect (i.e. ongoing telephone or online support), which may likely be cost-effective (Dickinson et al., 2017; McKechnie et al., 2014b). Indeed, the latter has been supported in a case study with a person with DLB and their care partner, where a multicomponent intervention, with embedded care partner education for 32 weekly sessions of one hour, reduced the care partner's distress and agitation in the person with DLB (Huh, Arean, Bornfield, & Elite-Marcandonatou, 2008). This suggests that

multicomponent approaches and care partner education, training and support are important and can lead to positive outcomes for both members of the dyad (Brodaty & Arasaratnam, 2012; Connors et al., 2018).

Interventions in PD have mostly focused on people with the condition and include a component for care partners embedded within the primary intervention. However, dyadic interventions or interventions for only the care partners have been less common (Hempel, Norman, Golder, Aguiar-Ibanez & Eastwood, 2008). From the interventions specifically offered to care partners, cognitive behavioural therapy, support groups and educational programs have been trialled and have been mostly based in clinics or institutional settings (Hempel et al., 2008; Mosley et al., 2017). Due to the small sample sizes, lack of cost-effectiveness evaluation and poor research designs, Hempel and colleagues (2008) concluded that there was lack of evidence to suggest the most effective and cost-effective interventions; however, some have appeared to result in positive outcomes for either care partners or both members of the dyad. Some evidence exists in support of educational programs, cognitive behavioural therapy and multidisciplinary interventions for care partners of people with PD, which have resulted in reduction of burden (Mosley et al., 2017). Despite the evidence-base of psychosocial interventions in dementia and PD, there remains a gap in knowledge about whether these can improve care partner outcomes long-term (Zarit, 2018) and whether they are effective among care partners of people with PDD (Hindle et al., 2013) and DLB (Connors et al., 2018).

Many of the aforementioned interventions are readily available for care partners of people with dementia and PD. However, most psychosocial interventions have not been specifically adapted to suit the needs of people with PRD, such as mobility, neuropsychiatric symptoms (e.g. apathy, anxiety, delusions) and impulse control behaviour (e.g. punding), and to suit the needs of care partners, such as feelings of tiredness, fatigue and burden due to care provision (McCormick et al., 2017a). Moreover, many dyads may not be aware that these interventions exist and thus may lack information how to access these interventions. Therefore, it is crucial that these interventions are adapted for PRD and that investigators and healthcare professionals

(i.e. GP, Parkinson's nurse specialist and consultants) work jointly to carry the field forward so that dyads can be informed and offered psychosocial interventions that could potentially benefit them.

To determine the effectiveness of psychosocial therapies for care partners of people with PRD, investigators can learn from previous interventions and consider adapting existing interventions to find a suitable, tailored and effective intervention for this population. For example, outside of PRD, interventions that included care partners as active participants have been found to improve behavioural and psychological symptoms in people with dementia as well as reducing care partners' depression levels and increased their quality of life (Brodaty & Arasaratnam, 2012; Leung, Orgeta, & Orrell, 2017).

Adapting an intervention for care partners of people with PRD is important for many reasons. First, the complex nature of care recipients' motor, cognitive and neuropsychiatric symptoms often requires continuous individual care. Second, care partners of people with PRD may experience higher rates of burden, stress, depression and anxiety than care partners of other types of dementia. Third, care partners of people with PRD may have to spend more time providing care and may not be able to leave their homes frequently due to the worry that something may happen to their partner; thus, they may not be able to travel to group sessions or take part in support groups. Therefore, taking into consideration the unique care partner profile within PRD, providing home-based interventions may be the most suitable option for this group.

To date, few interventions targeting cognitive and social functioning in people with PRD and their care partners have been tested and preliminary findings suggest that these can result in positive outcomes for both members of the dyad. Hindle and colleagues (2018) conducted a goal-oriented cognitive rehabilitation trial with 29 people with PRD, who received the intervention, and 26 care partners. The researchers found that care partners' quality of life and health status improved after people with PRD received the intervention (Hindle et al., 2018). Another study, which trialled a

dyadic cognitive training intervention with 223 people with PD-MCI and their care partners, found that care partners felt more relaxed and skilled to cope with various situations as a result of receiving an education programme (Reuter, Mehnert, Sammer, Oechsner & Engelhardt, 2012). Currently, the effectiveness of cognitive training in people with PDD and DLB and their care partners is unknown (Orgeta et al., 2015b). In an individualised Cognitive Stimulation Therapy (iCST) trial among people with dementia and their care partners (Orgeta et al., 2015a; Orrell et al., 2017), the care partners' quality of life as well as the perceptions of relationship quality held by the person with dementia improved.

Taking into consideration the dearth of psychosocial interventions for people with PRD and their care partners, our research team specifically adapted Cognitive Stimulation Therapy to meet the needs of this population (CST-PD, the INVEST trial; McCormick et al., 2017a), which was recently pilot tested (Leroi et al., under review; McCormick et al., 2017b; McCormick et al., in press). A comprehensive overview of the INVEST study is provided in Chapter 4 (Methods). Chapter 9 (Study 5) describes the evaluation of CST-PD with care partners.

2.3 Overview of intimate relationships in Parkinson's-related dementia

> *"Too often we underestimate the power of a touch, a smile, a kind word, a listening ear, an honest compliment, or the smallest act of caring, all of which have the potential to turn a life around."*
>
> *- Leo Buscaglia*

As human beings we long for social connections and interactions with others. Some of the relationships we form with other people may be romantic or intimate in nature, which is one of the most profound connections we form in our lives (Hendrick, 2004). Indeed, as stated in Baumeister and Leary's belongingness hypothesis (1995), the notion that we "need to belong is a fundamental human motivation" as "human beings have a pervasive drive to form and maintain at least a minimum quantity of lasting, positive, and significant interpersonal relationships" (p. 497). The will to find meaningful relationships is what drives people to seek partners with whom they can form, establish and maintain an intimate bond and connection leading to dating, short or long-term relationships, cohabitation and/or marriage. In fact, intimacy may be the reason why people marry (Schaefer & Olson, 1981) and marriage is considered to be one of the most intimate partnerships that grants affection, love and support to individuals (Levinger & Huston, 1990).

2.3.1 Terminology of intimate relationships

Many terms exist to refer to committed interpersonal relationships, such as romantic, intimate, marital, spousal or sexual relationships. Throughout this thesis, the terms '**intimate relationship'** and '**couple**' are used to indicate a committed dyadic partnership between the person with PRD and their partner. Taking into consideration that not every relationship may be romantic in nature, the term 'intimate' was preferred, which includes marital relationships as well as long-term, cohabiting, marriage-like intimate partnerships. Similarly, since not all couples are married, the

term '**life partner**' is used to denote a partner or a spouse who is providing care to their family member with PRD.

2.3.2 Components and importance of intimate relationships

2.3.2.1 Relationship quality

For decades a number of interrelated aspects within intimate dyadic relationships have been studied, including relationship quality, adjustment, stability, success, satisfaction (Spanier, 1979), and intimacy (Schaefer & Olson, 1981; Waring, 1981) as well as happiness, discord and well-being (Fincham & Bradbury, 1987; Proulx, Helms & Buehler, 2007). Early research into intimate interpersonal relationships has also focused on the concept of *love*, which comprises intimacy, passion and commitment, according to Sternberg (1986). However, most often, researchers have focused on assessing the quality of intimate relationships.

Relationship quality is a multifactorial construct and can be broken down into overall satisfaction, commitment, closeness or intimacy, passion, trust and love (Fletcher, Simpson, & Thomas, 2000). Spanier (1979, p. 290) defined relationship quality as "a subjective evaluation of a married couple's relationship with the range of evaluations constituting a continuum reflecting numerous characteristics of marital interaction and marital functioning". In the context of marriage, relationship quality encompasses adjustment, satisfaction, integration and happiness and can be seen in terms of its functionality and how the partners are affected by its functioning (Spanier, 1979). In fact, Spanier (1976) composed the Dyadic Adjustment Scale with similar components of relationship quality, which consists of *dyadic consensus* (degree to which partner agrees with the other), *dyadic satisfaction* (degree to which partner is satisfied with the relationship and is devoted to its continuance), *dyadic cohesion* (degree to which partners engage in mutual activities), and *affectional expression* (degree to which partner is satisfied with the level of affection and sex in the relationship). Spanier (1979) concluded that having a good relationship quality is important because it is

associated with well-being, adjustment, good communication, more happiness and higher relationship satisfaction, and could ultimately prolong a relationship (Rusbult, Martz, & Agnew, 1998).

2.3.2.2 Relationship satisfaction

Studies have increasingly explored relationship satisfaction, communication between partners, and specific types of intimacy, such as emotional and sexual intimacies (Basco, Prager, Pita, Tamir, & Stephens, 1992; Schaefer & Olson, 1981), which are interrelated (Yoo, Bartle-Haring, Day, & Gangamma, 2014). **Relationship satisfaction** has been defined in the context of interdependence theory (Kelley & Thibaut, 1978; Thibaut & Kelley, 1959), which sees the interaction between partners, dependence and satisfaction as the core elements in close intimate relationships (Rusbult & Buunk, 1993). The dyadic *interaction* consists of rewards (i.e. pleasure, enjoyment, fulfilment) as well as costs (i.e. stress, pain, shame) that each partner may receive in the relationship – the goal is to minimise costs and maximise rewards (Rusbult & Buunk, 1993). Specific components of a relationship, such as intimacy, love and disclosure may also be seen as rewards (Cherlin, 2004; Proulx et al., 1997). *Dependence* is the range of how much one needs a relationship or relies on the current relationship to obtain the outcomes one longs for, which could also revolve around the *quality of alternatives* which is the potential that the needs may be fulfilled outside of the current relationship (Rusbult et al., 1998). *Relationship satisfaction* is affected by the level of one partner fulfilling the most significant needs of the other partner (Rusbult et al., 1998). Each individual assesses the gains and benefits in their relationship as well as outputs they give to their partner. Relationship satisfaction is higher when the input-outcome ratio equates with that of the partner, whereas an imbalance in the ratio leads to dissatisfaction with the relationship (Rusbult & Buunk, 1993).

Relationship satisfaction can be enhanced and predicted by many constructs and can also determine the outcomes in one partner or both partners. Multiple studies have found that relationship satisfaction and relationship quality can contribute to mental or physical well-being, life satisfaction and happiness (Bookwala & Franks, 2005;

Falconier et al., 2015; Hassebrauck & Fehr, 2002; Heller, Watson & Ilies, 2004; Kiecolt-Glaser & Newton, 2001; Russell & Wells, 1994; Waldinger & Schulz, 2010); in contrast, low relationship quality can lead to poor mental health outcomes, such as depression (Baumeister & Leary, 1995; Beach, Jouriles, & O'Leary, 1985; Beach, Katz, Kim & Brody, 2003; Carr, Freedman, Cornman, & Schwarz, 2014; Clare et al., 2012; Levenson, Carstensen & Gottman, 1993; Proulx et al., 2007), higher levels of relationship instability and dissolution (Gottman & Levenson, 1992) and reduce couple cohesion and intimacy as well as increase aggression, criticism and blame (Beach et al., 1985). On one hand, longer relationship duration may decrease the association between relationship quality and well-being (Proulx et al., 2007), weaken relationship satisfaction (Karney & Bradbury, 1997; Kurdek, 1998; Vaillant & Vaillant, 1993) and reduce intimacy (Robinson & Blanton, 1993; Rowe & Meredith, 1982; Swensen, Eskew, & Kohlhepp, 1984). On the other hand, several factors can improve relationship satisfaction, for instance *overall intimacy* (Greeff & Malherbe, 2001; Merves-Okin, Amidon & Bernt, 1991; Robinson & Blanton, 1993; Schaefer & Olson, 1981; Toldstedt & Stokes, 1983; Waring, 1981), *sexual intimacy* (Byers, 2005), *communication* (Bradbury & Karney, 2013; Gottman, 1994; Lavner, Karney, & Bradbury 2016; Woodin, 2011), *commitment* (Rusbult et al., 1998) and *self-disclosure* (Hansen & Schuldt, 1984; Hendrick, 1981). Despite the general consensus linking relationship satisfaction and intimacy, it is difficult to determine the causal association between the constructs due to their interdependence and reciprocity (Byers, 2005). Therefore, it is imperative that each relationship component is evaluated independently with appropriate scales and is defined clearly.

Studies have explored what makes a long-term relationship satisfying and why people stay together, even during the difficult times. In Kaslow and Hammerschmidt's (1993) work, the following components were found to contribute to relationship satisfaction in couples who have been together for 25 to 45 years: (a) trust, fidelity, feeling safe, (b) good problem-solving and coping skills, (c) permanent commitment, (d) open, honest and good communication, (e) shared values, interests and activities, (f) a good balance between spending time together as well as apart, (g) mutual appreciation and reciprocity, and (h) deep love, friendship and continuing finding each other attractive.

Several of these aspects are what contribute to relationship satisfaction with top three being love, mutual trust and mutual respect (Kaslow & Robinson, 1996). People remain in their relationships for many reasons for example due to a belief that marriage is a partnership for life, a sense of responsibility toward the partner, an enjoyment of their established lifestyle, religious beliefs about the holiness of marriage, a sense and appreciation of closeness resulting from shared experiences, and comfort with each other (Kaslow & Robinson, 1996). The authors concluded that intrinsic factors (e.g. love and lifelong commitment) were the main aspects in satisfied couples and extrinsic factors (e.g. responsibility to partner and religious commitment) were the primary elements in couples who were dissatisfied with the relationship (Kaslow & Robinson, 1996).

2.3.2.3 Commitment

Commitment in relationships is another well-researched construct which is a central component in relationships (Kaslow & Robinson, 1996; Rhoades, Stanley, & Markman, 2010; Rusbult, 1980; Stanley & Markman, 1992). **Commitment** is "a long-term orientation, including feelings of attachment to a partner and desire to maintain a relationship, for better or worse" (Rusbult & Buunk, 1993, p. 180). Couples may experience challenges in the face of conflicts, disagreements and differences of opinion and, in turn, may doubt in the maintenance of the relationship. Thus, it is important that both partners communicate their desired outcomes to each other. Generally, couples who adapt to each other's differences, work closely towards mutual goals and adjust to changes in their relationship report stronger commitment (Robinson & Blanton, 1993).

Stanley and Markman (1992) have proposed that commitment is comprised of dedication as well as constraints which are further divided into three specific commitment types. *Dedication* is the desire and motive to build the quality of the relationship and maintain it in the future, whereas constraint commitment can be: (a) *perceived constraints*, which is the societal pressure to stay together or negative effect on partner(s) if the relationship terminates, (b) *material constraints*, which refers to

mutual investments such as owning a pet, planning a holiday together, sharing debt, and (c) *felt constraints*, which are the internal or external pressures (such as investments made) to an individual who can feel trapped or stuck due to the feeling that they need to stay in the relationship (Rhoades et al., 2010; Stanley & Markman, 1992). These four aspects of commitment were both associated with and predicted relationship stability (Rhoades et al., 2010). The authors discussed that the most integral part of intimate relationships is the knowing that there is a future with one's partner which is based on the wish for a future (dedication) and on the aspects that can strengthen relationship continuance (constraints) (Rhoades et al., 2010). Thus, it is important to take commitment into consideration when exploring intimate relationships.

2.3.2.4 Intimacy

A wealth of studies has focused on researching intimacy but some confusion exists between the definitions of intimacy. Generally, **intimacy** is defined as a multifaceted, dynamic construct encompassing a person's subjective experience of closeness, connectedness and commitment with one's romantic partner, which arises from dyadic processes involving self-disclosure, communication, acceptance, affection, empathy, mutual trust and validation (Hook, Gerstein, Detterich, & Gridley, 2003; Laurenceau, Feldman Barrett, & Rovine, 2005; Moss & Schwebel, 1993; Schaefer & Olson, 1981; Sternberg, 1986, 1987; Waring, 1984; Wynne & Wynne, 1986). Intimacy has been difficult to extinguish from self-disclosure as they share many similarities but studies suggest that intimacy is predicted by self-disclosure (Laurenceau et al., 2005; Schaefer & Olson, 1981; Waring, 1981). *Self-disclosure* (or cognitive self-disclosure) means verbally communicating personal information to the other person, such as our emotions, thoughts, beliefs and attitudes as well as forming our self-awareness (Waring, 1981) and is described by mutual reciprocity (Schaefer & Olson, 1981). Intimacy has also been defined as an amalgamation of *affection* (expression of emotional closeness), *compatibility* (ability to work and play together), *sexuality* (fulfilment of sexual needs), *cohesion* (commitment to the relationship), *conflict resolution* (ease of resolving differences of opinion), *autonomy* (positive

connectedness to family and friends), *expressiveness* (communicating one's thoughts and beliefs to other) and *identity* (couple's level of self-confidence and self-esteem), which are united by communication within the relationship (Waring, 1981).

Another widely used definition of **intimacy** originates from Schaefer and Olson (1981), who proposed that intimacy is both a process and an experience which arises from sharing an intimate experience together and disclosing personal information to each other. Intimacy can differ between men and women. For women, intimacy can lead to higher satisfaction with the relationship and more happiness, whereas for men intimacy can be transferred onto different areas of daily life functioning (Greeff & Malherbe, 2001; Reichman, 1989). Intimacy is often assessed as a multidimensional construct taking into consideration five types of intimacies: (1) emotional – perceived closeness of feelings, (2) intellectual – extent of sharing thoughts and ideas, (3) social – having mutual friends, (4) recreational – common interests and hobbies, and (5) sexual – sharing physical closeness, affection and/or sexual activity, which are measured with the 'Personal Assessment of Intimacy in Relationships' scale (Schaefer & Olson, 1981).

2.3.2.5 Communication

Key elements in a committed interpersonal relationship are verbal and non-verbal **communication.** Communication is closely related to relationship satisfaction, intimacy and self-disclosure, and is "the primary vehicle through which we define our relationships" (Fitzpatrick & Best, 1979, p. 167). Effective communication between partners, where couples can share, open up, discuss thoughts and concerns and validate each other's self-disclosure, can increase intimacy (Laurenceau et al., 2005; Mitchell et al., 2008; Yoo et al., 2014). In contrast, unfriendly, negative and demanding communication can lead to lower relationship satisfaction (Gottman & Notarius, 2000). A couple may experience challenging situations which can lead to disagreements and conflicts. Couples who report higher relationship satisfaction are more willing to discuss and solve conflict and have more effective communication than couples who are distressed or dissatisfied with the relationship, who may avoid resolving conflict (Bradbury & Karney, 2013; Gottman, 1994; Kaslow & Robinson, 1996; Lavner et al.,

2016; Woodin, 2011). Kaslow and Robinson (1996) found that honesty was the primary component that improved the quality of communication. Communication is part of the intimacy process as one partner (i.e. speaker) communicates personal information to the other (i.e. listener), who responds in a supportive, understanding and empathic manner. In order for this relationship to be intimate, it is important that the speaker perceives the listener's responsiveness as accepting, validating and caring (Reis & Shaver, 1988). Two-way communication is central in a dyadic relationship and it can be seen as a 'doorway' to intimacy, as talking, sharing, listening, trusting as well as respecting each other can increase closeness, connectedness, intimacy and, subsequently, relationship satisfaction and quality.

2.3.2.6 Loneliness

One important reason why people want and need social and intimate relationships is to avoid feelings of loneliness. Older adults may be particularly at risk of being socially isolated and/or lonely as they may have become a widower, are more geographically mobile, live on their own and may have friends who passed away (Valtorta & Hanratty, 2012). **Loneliness** is a perceived dissatisfaction of the quantity and quality of one's current relationships or the difference between the current and desired amount of social contact (Hawkley & Cacioppo, 2010; Ong, Uchino, & Wethington, 2016; Peplau & Perlman, 1982; Pinquart & Sörensen, 2001; Steptoe, Shankar, Demakakos, & Wardle, 2013; Wheeler, Reis, & Nezlek, 1983). *Social isolation* is lack of (or minimal) social contact and reduction of social network size, which is quantifiable and therefore objective, whereas *loneliness* is a subjective perception of longing for close and emotional relationships with others (Ong et al., 2016; Steptoe et al., 2013). People who may be socially isolated and lonely have a risk of health deterioration, developing physical, psychiatric or cognitive illnesses (including dementia), and mortality (Hawkley & Cacioppo, 2010; Holt-Lunstad, Smith, Baker, Harris, & Stephenson, 2015; Ong et al., 2016; Steptoe et al., 2013). Loneliness can also contribute to lower quality of life, less satisfaction with life and depression (Gerino, Rolle, Sechi, & Brustia, 2017; Singh & Misra, 2009). Thus, loneliness is an important public health concern, particularly in later life (Gerst-Emerson & Jayawardhana, 2015).

It is also possible for loneliness to emerge in married couples. Individuals experienced more emotional and social loneliness when one partner had health problems, received little or no support from the partner, no longer had frequent conversations or had more arguments, and had an unpleasant or non-existent sexual life (de Jong Gierveld, van Groenou, Hoogendoorn, & Smit, 2009). However, people who were married and had frequent contact with friends reported less loneliness (Pinquart & Sörensen, 2003b). These findings highlight that commitment, ability to resolve conflict and good communication can strengthen a partnership, which could potentially diminish loneliness in partners and ultimately lead to better outcomes for both individuals.

2.3.3 Intimate relationships in Parkinson's-related dementia

Research into long-lasting intimate relationships with older people has increased over recent decades. As people live longer, many couples celebrate their 50th wedding anniversaries (Melton, Hersen, Van Sickle, & Van Hasselt, 1995). When studying long-term relationships, several factors need to be considered. For instance, in Kaslow and Robinson's (1996) work the following qualities were highlighted: the duration of the relationship or marriage; presence of a long-term illness or neurodegenerative condition; couples' ability to overcome conflicts and disagreements; will to maintain the relationship, and presence or absence of relationship satisfaction, relationship quality, good communication, intimacy and commitment. One partner's acute or chronic physical or mental ill health poses challenges for the couple and can lead to instability in the relationship, increased costs and reduced rewards for life partners (Melton et al., 1995). It could also lead to role change and additional responsibilities for life partners (Boylstein & Hayes, 2012; Evans & Lee, 2014; Holdsworth & McCabe, 2018; Martin, 2016; Pozzebon, Douglas & Ames, 2016; Quinn, Clare, & Woods, 2009).

A neurodegenerative condition, such as PRD, can challenge the couple even more than other diseases because it is incurable and will continue to progress over time. Thus, efficient and effective coping strategies are required to overcome these challenges and sustain relationships, because lack of these strategies can lead to increased burden

and health issues in the care partner, institutionalisation of the person with PRD and eventually relationship breakdown (Wright, 1998). Since the majority of studies in this field have focused on exploring relationships in dementia and in PD, these findings are presented below and the potential relevance to PDD and DLB is provided.

Both dementia and PD have a profound effect on the person, the care partner and their relationship (Hodgson, Garcia, & Tyndall, 2004; Martin, 2016). People with PD have reported significant reduction in sexual functions, although the non-sexual relationship aspects, for example talking about one's feelings or tenderness, increased with the duration of the disease (Buhmann et al., 2017). Men with PD tend to withdraw from the relationship, may have had increased thoughts of divorce and may have reported dissatisfaction with the relationship and sexuality since the onset of PD, more so than women with PD (Buhmann et al., 2017). Mutuality, defined as the positive quality of a partnership consisting of love and affection, reciprocity, shared values and shared pleasurable activities (Archbold, Stewart, Greenlick, & Harvath, 1990), remains relatively high at mild to moderate stages of PD but can be significantly lower at an advanced stage of PD (Carter et al., 2008). Likewise, in another study, both partners' mutuality levels were similar but people with PD reported higher reciprocity than their partners (Karlstedt, Fereshtehnejad, Aarsland, & Lökk, 2017). Mutuality, alongside with non-motor symptoms, was also found to be a predictor of health-related quality of life for people with PD, whereas mutuality and cognition were the main predictors of burden in life partners (Karlstedt et al., 2017). In a study with people with early onset PD (i.e. less than 50 years of age) and their partners, both relationship and sexual dissatisfaction were common and these levels were similar in both members of the couple (Wielinski, Varpness, Erickson-Davis, Paraschos, & Parashos, 2010). These studies highlight that the impact of PD on the couple is substantial and research should look into addressing these issues through dyadic interventions to improve outcomes for both partners.

The advancing nature of dementia increases the person's memory loss, confusion, agitation and inability to communicate, which may lead them to not recognising one's partner and forgetting that they are married (Evans & Lee, 2014). As a consequence,

the life partner might start to doubt whether the marriage still exists (Evans & Lee, 2014), which can be applicable in PRD as well. Thus, the central theme describing relationships within dementia is often 'loss' – loss of a person, loss of relationship, mutual companionship and connectedness (Evans & Lee, 2014; Pozzebon et al., 2016). Quinn and colleagues (2009) found in their systematic review that the relationship with the person with dementia had changed or was lost, and reciprocity, affection, relationship quality, intimacy and dyadic communication were diminished for life partners, despite spending more time together. Similarly, life partners of people with PD experienced feelings of loss and helplessness and felt overwhelmed and unable to cope with the cognitive impairment of the care recipient (Lawson et al., 2018). As early as the mild cognitive impairment stage, communication is said to reduce, leading to lower marital satisfaction in life partners (Garand et al., 2007) and a greater decline in satisfaction when dementia had emerged (Davies et al., 2010). Although the majority of people in previous studies had Alzheimer's dementia or vascular dementia, these findings could potentially apply to people with PDD and DLB as well; however, due to the limited number of studies in PRD, it is difficult to draw conclusions among people with PRD and the life partners.

The preceding findings resonate with those found by Holdsworth and McCabe (2018) who further added that changes in identity, self-esteem, commitment as well as sexual activity and satisfaction were important determinants of relationships. Indeed, sexuality remains important at later age, despite reduced sexual activity both in MCI and dementia due to motor inabilities (Davies et al., 2010). Sexuality can also contribute to quality of life and well-being in older adults and life partners of people with AD (Davies, Sridhar, Newkirk, Beaudreau, & O'Hara, 2012; Flynn & Gow, 2015). Thus, future studies should explore relationships, intimacy and sexuality and do so from three angles: viewed from the person with dementia, their partner, and the couple as a dyad (Holdsworth & McCabe, 2018). Such research is currently lacking with people with PRD and their partners.

Several specific factors have been explored in terms of relationship satisfaction in dementia. Studies have determined that pre-dementia relationship satisfaction is

important as it is associated with less care partner burden, less reactivity to partners' symptoms and behaviour, better communication, and problem solving skills (Steadman et al., 2007) as well as higher quality of life, higher caregiving satisfaction, less stress and depression in care partners (Kramer, 1993; Morris, Morris, & Britton, 1988). At the time when a neurodegenerative condition, such as dementia or PD, had emerged some life partners did not report a change in their relationship or closeness (Martin, 2016), and some even felt closer to the partner than before (de Vugt et al., 2003; Martin, 2016) and reported higher intimacy (Shavit, Ben-ze'ev & Doron, 2017). However, another study showed that people with dementia reported higher relationship quality compared to their partners (Wright, 1991). These contrasting findings suggest that it is important to include both members of the dyad when exploring relationships so that comparisons of relationship satisfaction and quality between partners could be made, which has been undertaken in Study 4 in this thesis.

Having a close relationship with one's partner can be protective. More satisfaction with intimacy was associated with less stress and fewer depressive symptoms, particularly in female care partners (Davies et al., 2012). In PD, higher mutuality was related to better mental health outcomes for partners, lower PD severity as well as lower burden and higher quality of life in the care partner (Tanji et al., 2008). The ability to remain positive when having PD or living with a care recipient who has PD has been found to contribute to higher marital quality for the couple (Mavandadi et al., 2014). These findings resonate with Habermann's (2000) study who stated that PD affected couples' closeness and communication positively. Despite these encouraging findings, PD has been found to have a detrimental effect on the relationship and lead to poor marital adjustment (Carter & Carter, 1994). Thus, further research is required to explore the consequences of PD and PRD on the person, life partner and their relationship.

Researchers have also identified the elements that hold the relationship together in dementia. Many couples saw that 'quid pro quo', commitment, relational bond, spirituality, and reaching out for emotional support were important components why life partners stayed with their partner with dementia (Loboprahbu, Molinari,

Arlinghaus, Barr, & Lomax, 2005). Many of these concepts are similar to the ones portrayed by Kaslow and Hammerschmidt (1993) and Kaslow and Robinson (1996) (see Section 2.3.2.2), which also illustrates the universality of committed intimate relationships even in the case of dementia. Importantly, being married (Hakansson et al., 2009; Xu, Thomas, & Umberson, 2016) and having higher relationship closeness can prevent cognitive impairment in later life or slow down the progression of cognition and functional abilities in the person with dementia (Norton et al., 2009).

When one partner is diagnosed with dementia, the couple goes through a variety of changes. Kaplan (2001) characterised couplehood transitions within AD on a "We" and "I" continuum whereby five different groups were found: (A) "Til death do us parts", (B) "We, but…", (C) "Husbandless wives/Wifeless husbands", (D) "Becoming an I", and (E) "Unmarried marrieds". These five groups differed in terms of their commitment, status, coping and future outlook. For example, group A only saw themselves as 'We', whereas group C felt their partner (and therefore their relationship) was not the same, and group E, although legally married, did not feel as if they were married. Furthermore, groups C, D and E felt more isolated and were starting to re-establish themselves as individuals again rather than seeing themselves as a couple (Kaplan, 2001). Similarly, Shavit and colleagues (2017) studied the changes of the relationship after the emergence of dementia and found five types of changes in love: love died, love became weaker, love did not change, love was enhanced and the partner fell in love again. According to Kaplan's typology of couplehood, it could be proposed that for group A love did not change or was enhanced, for groups B and C love became weaker, and for groups D and E love died. Moreover, Kaplan (2001) suggested that if people with dementia have been admitted to the care home, life partners of groups D and E may start to look for new relationships and may fall in love again.

There is a growing body of evidence suggesting that both dementia and PD can significantly impact relationships, but it is important to consider relationship changes specifically in PDD and DLB, which have been underexplored. Notably, PRD can increase burden and mental health issues in life partners, as described in section 2.2.4 (Care provision in PRD). Research outside of PD shows that one partners' depression

can contribute to relationship dissatisfaction, lower levels of communication and problem-solving abilities as well as difficulties maintaining intimacy (Basco et al., 1992). In turn, higher loss of intimacy can lead to higher levels of depression (Morris et al., 1988). Similarly, lower marital quality in people with PD can contribute to higher anxiety in life partners (Mavandadi et al., 2014). In cognitively intact people with PD, the motor symptoms had a significant impact on the relationship (Tanji et al., 2008) but when cognitive decline had emerged, non-motor symptoms were the most prominent stressors on couples' relationships (Karlstedt et al., 2017). However, to my knowledge, no study has explored relationship changes among people with PD-MCI, PDD and DLB and their life partners collectively, which is a gap in knowledge. Importantly, evidence in studies with people with dementia and PD suggests that couples may experience both positive and negative effects on their relationships as a result of the neurodegenerative condition; therefore, it is crucial to conduct studies in Parkinson's-related dementia.

2.3.4 Psychosocial interventions to improve relationship satisfaction

An increased understanding of how relationships change as a result of a long-term health condition or serious illness (including dementia and PD) has prompted researchers to develop interventions to address these changes and improve couples' relationship. Frequently, the psychosocial interventions are focused on enhancing care partner outcomes, such as burden, stress, mental health, quality of life, social support and relationship quality (Abrahams et al., 2018; Gilhooly et al., 2016; Hindle et al., 2018; Hopwood et al., 2018; Kwon, Ahn, Kim, & Park, 2017; Laver, Milte, Dyer, & Crotty, 2017; McKechnie et al., 2014a, 2014b; Orrell et al., 2017). However, only a handful of studies have focused on improving dyadic outcomes for both partners such as mutual interaction and strengthening relationship satisfaction and quality. Furthermore, many interventional studies have exclusively targeted individual outcomes and not taken into account that partners' outcomes are interrelated and should be studied jointly to understand the bidirectional effects on each partners' outcomes, such as health, well-being and relationship satisfaction (Mavandadi et al., 2014; Van't Leven et al., 2013). Davies and colleagues (2010) recommended that

interventions should be provided already at the MCI stage to help couples adjust to changes and behaviours and modify activities and expectations about the future of their relationship. Already a decade ago, researchers acknowledged that interventions should address improving the mutuality, interaction and relationship quality of people with PD and their partners which could potentially decrease strain in the care partner and improve their mental health (Tanji et al., 2008). Dyadic interventions could potentially help to sustain relationships, maintain quality of life, reduce burden and delay institutionalisation, which subsequently can reduce costs in the health and social care system (Davies et al., 2010).

Couple-centred interventions trialled outside of dementia and PD have found some positive findings. Dyadic marital interventions, such as behavioural couple therapy, partner-assisted interventions and disorder-specific interventions, are beneficial in treating depression and substance use disorders and could be more effective than individual interventions, as has been posited by Whisman and Baucom (2012). A dyadic group-based intervention, aimed at relationship skills training for people with brain injury and their partners, resulted in improved relationship satisfaction and quality as well as lower negative communication for partners (Backhaus et al., 2016).

Interventions have also found that improving couples' communication skills could enhance relationships and reduce distress; however, these can only be effective and suitable when poor communication is the cause of marital distress (Lavner et al., 2016). In cancer, dyadic interventions have been beneficial in enhancing couple communication, psychological distress and relationship functioning (Regan et al., 2012). Another review found that enhancing and promoting communication, problem solving, self-disclosure, responding in an empathic way and providing sexual education and counselling could help restore intimacy and support family connections (Kardan-Souraki, Hamzehgardeshi, Asadpour, Mohammadpour, & Khani, 2016).

Several dyadic psychosocial interventions in dementia have been developed addressing relationship satisfaction and quality in both members of the dyad. One such intervention, called 'The Couples Life Story Approach', was specifically developed to

improve marital relationship quality as well as quality of life for the person with dementia and their spouse (Ingersoll-Dayton et al., 2013). The primary component of the intervention was life review, which includes a process of recalling, organising and evaluating one's life, which in turn can promote purposeful and reciprocal engagement in the couple (Ingersoll-Dayton et al., 2013; Kwak, Han, & Ha, 2018). The pilot trial of the intervention demonstrated the feasibility and acceptability of the intervention and also appeared to increase intimacy, mutuality and couplehood for the partners (Ingersoll-Dayton et al., 2013). Another study trialling this intervention with 102 Korean couples, where one partner had AD, found that doing an enjoyable mutual activity such as life review increased joy, reminiscence, communication and relationship quality for some of the couples (Kwak et al., 2018), suggesting the participation of both partners could be beneficial for the couple.

For the person with dementia, quality of life can be improved by supporting relationships with care recipients and encouraging social participation (Martyr et al., 2018), which can also apply to life partners and can be addressed by providing support to both partners at once. Despite the aforementioned promising results, Bielsten and Hellström (2017a; 2017b) have argued that many couple-based interventions in dementia have not considered the nature and the quality of the relationship. Furthermore, the authors concluded that many studies did not have a dyadic approach, did not consider the views of people with dementia, did not tailor support and had a negative outlook of outcomes, highlighting that the components of the relationship should not be disregarded in couple-centred interventions (Bielsten & Hellström; 2017a; 2017b).

2.3.5 Application of the theory in the thesis

There are currently no interventions that have been developed to improve relationship satisfaction and quality among people with PD-MCI, PDD and DLB and their life partners. The current literature review suggests, however, that dyadic interventions can be beneficial in improving specific relationship aspects, such as interaction, mutuality, relationship satisfaction and quality, among people with dementia and their

partners; thus, it is hypothesised that a dyadic intervention may be beneficial among life partners of people with PRD. One intervention that improved the relationship quality between the person with dementia and their care partner was individual Cognitive Stimulation Therapy (Orgeta et al., 2015a; Orrell et al., 2017). It could be argued that the reason why relationship quality improved is that the couple took time to sit down, reconnect, interact and communicate, which many couples may not have done due to loss of communication by the person with dementia, changes in the relationship and shift into a care provider-care recipient roles. Thus, it is likely that an appropriately adapted and tailored iCST could be suitable for people with PRD and their care partners to improve their outcomes.

Our research team (I.L., S.V., K.R.M., S.A.M.) specifically adapted Cognitive Stimulation Therapy for people with PRD (CST-PD, the INVEST trial; McCormick et al., 2017a; see Chapter 4: Method) and recently completed a pilot trial with dyads of people with PRD and their care partners. The primary aim of the INVEST study was to evaluate operational aspects (i.e. feasibility of recruitment, acceptability and tolerability of the intervention) and the efficacy of CST-PD for people with PRD (i.e. cognitive impairment and quality of life), which will provide the necessary information for a subsequent full-scale RCT trial (McCormick et al., 2017b). However, taken into account the benefits that life partner participation in dyadic interventions can have (Leung et al., 2017), it was important to also evaluate the effects of CST-PD on life partners in terms of relationship satisfaction, burden and mental health, which were undertaken in Study 5 (Chapter 9).

CHAPTER 3: Aims of the thesis

The overarching aim of this thesis was to explore the impact of mild cognitive impairment (PD-MCI) or dementia (PDD) in Parkinson's disease and dementia with Lewy bodies (DLB)[2] on the outcomes of life partners, such as relationship satisfaction, burden and stress, physical and mental health, quality of life and feelings related to care provision. The objective was to gain a thorough understanding of the changes that life partners experienced as a result of the neurodegenerative condition of the care recipient via the application of quantitative and qualitative methods.

The specific aims of the studies were as follows:

- To describe the sociodemographic profile of life partners of people with PRD, compare clinical outcomes of life partners according to the clinical syndrome (PD-MCI, PDD or DLB), and evaluate psychometric properties of the scales to provide recommendations for future studies (Study 1);
- To investigate the factor structure of the Zarit Burden Interview (Zarit, Reever, & Bach-Peterson, 1980) in life partners of people with PRD and examine the associations and predictors between the emerging factors and the demographic and clinical features (Study 2);
- To explore changes in long-term intimate relationships in PD-MCI, PDD and DLB through the perspective of caregiving life partners (Study 3);
- To examine the associations between depression, anxiety, quality of life and relationship satisfaction among people with PD-MCI, PDD or DLB and their life partners and explore actor and partner effects (Study 4);
- To conduct a secondary analysis of a pilot randomised controlled trial of Cognitive Stimulation Therapy in Parkinson's-related dementia (CST-PD) to explore whether relationship satisfaction, burden, quality of life and mental health of life partners improved as a result of doing CST-PD compared to the control group.

[2] PD-MCI, PDD and DLB are collectively referred to as Parkinson's-related dementia (PRD) throughout this thesis.

CHAPTER 4: Methods

The full protocol of the INVEST study has been published:

McCormick, S. A., McDonald, K. R., **Vatter, S.,** Orgeta, V., Poliakoff, E., Smith, S. J., Silverdale, M. A., Fu, B., & Leroi, I. (2017b). Psychosocial therapy for Parkinson's-related dementia: study protocol for the INVEST randomised controlled trial. *BMJ Open, 7(6)*, e016801.

This chapter provides a detailed overview of the quantitative methods used in the following studies:

- **Study 1** is a cross-sectional study with life partners of people with Parkinson's-related dementia (PRD);
- **Study 2** is a factor analysis of the Zarit Burden Interview (ZBI; Zarit et al., 1980) with life partners of people with PRD;
- **Study 4** is a cross-sectional dyadic analysis study which applied an actor-partner interdependence model (APIM) with couples where one partner has PRD;
- **Study 5** is a secondary analysis of a pilot feasibility randomised controlled trial (RCT) of the Cognitive Stimulation Therapy in Parkinson's-related dementia (CST-PD) with life partners.

Study 3, a qualitative study, is described in detail in Chapter 7.

The studies described here are nested within the INVEST study, which was an exploratory single-blind two-arm pilot and feasibility RCT with 76 participant-dyads. Dyads were randomly allocated either to the Cognitive Stimulation Therapy in Parkinson's-related dementia (CST-PD) arm or treatment as usual (TAU) arm. All dyads consented once to the INVEST study; therefore, there is one ethical approval for all studies (See Section 4.2.8, Appendix A). Additionally, a sub-sample of life partners of people with PRD was recruited for Studies 1 and 2 via a postal questionnaire, for which ethical approval was granted via an amendment to the INVEST study.

The methods of the four quantitative studies (i.e. design, inclusion and exclusion criteria of participants, recruitment, sample size, power calculations, procedure, measures and ethical considerations) are described in Section 4.2. The specific aims and objectives of each study are provided in individual study chapters. The current chapter follows the Standard Protocol Items Recommendations for Interventional Trials guidelines (SPIRIT, Chan et al., 2013) to maximise clarity and transparency of the methods section.

My role in the INVEST study as an unblinded researcher was to liaise with clinicians, nurses and researchers to identify potential participants, screen and recruit participant-dyads, randomise participant-dyads, conduct informed consent visits and therapy training visits with participant-dyads, and undertake weekly phone calls. I was also responsible for the day-to-day coordination of the INVEST study (i.e. ensuring ongoing recruitment of participants, providing up-to-date trial documentation, etc) and supporting the blinded and unblinded researchers at all sites throughout the trial.

4.1 Patient and Public Involvement

In all stages of developing the CST-PD intervention, the patient and public involvement (PPI) representatives (i.e. care recipients and care partners) were included to ensure their feedback was taken into account during the design process (McCormick et al., 2017a). INVOLVE, a national advisory group, is part of and funded by the National Institute of Health Research (NIHR) who encourage the support of active public involvement in NHS as well as public health and social care research (INVOLVE, 2018). PPI is defined as "research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them" (INVOLVE, 2012, p. 6). The INVEST study team continued to work closely with two couples (both involved a person with PD and their life partner) throughout the study. The PPI representatives contributed to the PhD studies by commenting on the language use in study-related documentation, providing feedback on the interview schedule, actively participating in study steering committee meetings, promoting the study through their networks and disseminating the research.

The importance of including members of the public in the conduct of health and social care research studies and interventions has been emphasised by the NIHR (2014, 2017), INVOLVE (2012), Department of Health (2004), the NHS plan (Department of Health, 2000), NICE (2013) and WHO (2016) and is regarded a good practice. PPI includes active collaboration between researchers and people with a specific condition for whom a certain study, intervention or a drug may be developed for and/or members of the public (Thompson, 2007). The aim of including PPI in research is to plan, design, guide, conduct and circulate research studies (Howard Wilsher, Brainard, Loke & Salter, 2017) and maximise the applicability, tolerability, transparency and adherence of a particular intervention so that it meets the needs of the people for whom the intervention is targeted for.

4.2 Design

4.2.1 Pilot and feasibility study design

Several funding bodies, such as NIHR and Medical Research Council (MRC, Craig et al., 2008) have suggested undertaking pilot or feasibility studies prior to fully powered RCTs; however, the understanding of the definitions and differences between '*pilot*' and '*feasibility*' is not fully clear and should be explained in more detail. The MRC framework (Craig et al., 2008) for developing and evaluating complex interventions recommended running a 'feasibility and piloting' stage after the intervention development phase which consists of conducting operational procedures to observe acceptability, recruitment and retention of participants and to calculate sample sizes for a future study. Furthermore, the MRC stated that a pilot study should focus on the concerns from the development stage and implement both qualitative and quantitative methods in addressing potential barriers and issues (Craig et al., 2008). In contrast, the NIHR (2017) has separately defined feasibility and pilot studies: (1) feasibility studies seek to determine whether the study can be done through exploration of various parameters that are required for the main RCT (e.g. willingness

of participants to be randomised, number of participants, rates of follow-up, response rates and adherence, assessing the outcome measure, etc.) and, (2) pilot studies involve the undertaking of a smaller version of the definitive trial to test whether the components of the main study, such as recruitment, randomisation, treatment and follow-up assessments, can work together.

Recently, Eldridge and colleagues (2016a) undertook an in-depth study to develop a framework using the Delphi process, involving a review of pilot and feasibility studies, and discussion with experts in a consensus meeting and at a methodology conference. The authors concluded that a feasibility study explores whether a specific aspect of a research project can be done and how it can be done, whereas a pilot study focuses on testing a specific research design on a smaller scale ahead of the main definitive RCT (Eldridge et al., 2016a); this resembles the NIHR definition (2017). The PhD studies are nested within the INVEST study, which was a pilot and feasibility study of CST-PD with an embedded process evaluation. The primary aim of the INVEST study was to evaluate the processes of study components (e.g. recruitment, randomisation, intervention, assessments) and tolerability of the intervention by participating dyads (*feasibility*) (McCormick et al., 2017b; McCormick et al., in press). The secondary aim was to explore the outcome measures for people with PRD and their care partners (*pilot*; Leroi et al., under review). Since the PhD studies focus exclusively on life partners (with the exception of Study 4 which conducted a dyadic analysis with couples), the studies in this thesis will not focus on the operational and practical elements of the INVEST study; rather, the quantitative studies (Studies 1, 2, 4 and 5) will explore specific outcomes for life partners of people with PRD via multiple analyses.

4.2.2 Participants

The INVEST study recruited participant-dyads. Participants with a diagnosis of Parkinson's disease and mild cognitive impairment (PD-MCI) or dementia (PDD), or dementia with Lewy bodies (DLB) (collectively referred to as Parkinson's-related dementia, PRD), and their care partners, were recruited to the INVEST study. A care

partner could have been a family member, relative, friend, paid carer or a personal consultee. For the purposes of the quantitative studies presented in this thesis, only married or co-habiting couples and/or life partners of people with PRD were included. An additional postal questionnaire was conducted (Studies 1 and 2) to recruit life partners of people with PRD.

4.2.2.1 Inclusion and exclusion criteria of participants

The inclusion and exclusion criteria of participant-dyads in the INVEST study and in the PhD studies is provided in Table 4.1.

4.2.2.2 Sample size and power calculations

The INVEST study recruited 76 participant-dyads. As the INVEST study was an exploratory pilot study, it was not intended to be fully powered as the primary aim was to assess operational aspects (i.e. recruitment, retention, feasibility) and the secondary aim was to explore the efficacy of CST-PD for people with PRD. Relying on the sample size guidance for pilot exploratory studies (Browne, 1995; Julious, 2005; Whitehead, Julious, Cooper & Campbell, 2016), a conservative approach was taken whereby the anticipated standardised effect size for the INVEST study was 0.4 with a desired power of 80% and an assumed correlation coefficient 0.5 between baseline and follow-up outcomes. This resulted in a sample size of 27 completers per randomisation arm and required 38 dyads per arm to allow for 30% attrition rate.

Since the studies of this PhD were nested in the INVEST study and were exploratory in nature, no power calculations prior to the analyses were performed (Jones, Carley, & Harrison, 2003), and the sample sizes for each quantitative study were not pre-determined. The number of life partners resulted from a pragmatic decision to include all eligible participants from the INVEST study (n = 57) in the PhD studies. The sample size for the postal questionnaire survey was also not predetermined; however, a minimum of 100 participants was required for the factor analysis study (Study 2) (Gorsuch 1983; Kline, 1994).

Table 4.1 Inclusion and exclusion criteria of participant-dyads.

	Included if:	Excluded if:
People with PRD	• Must have received a diagnosis of probable PD-MCI, PDD or DLB based on standard clinical diagnostic criteria (Emre et al, 2007; Litvan et al., 2012; McKeith et al., 2005; Appendix B, Table B.1), which is determined by the referring clinician; • Must have been willing and well enough to participate in 20 – 30 minute sessions of CST-PD, two or three times per week; • Must have been stable on medication regime four weeks prior to study entry.	• Unwilling or not well enough to participate in 20 – 30 minute sessions of CST-PD, two or three times per week; • No care partner or the contact with the care partner was < 3 times a week; • Living in a residential care; • Had a severe physical illness; • Could not understand English or were non-literate; • Were taking part in another dementia intervention research project at the same time.
Care partners	• Provided care to a person with PD-MCI, PDD or DLB; • Willing and well enough to deliver 20 – 30 minute sessions of CST-PD, two or three times per week.	• Did not provide care to a person with PD-MCI, PDD or DLB; • Had a severe physical illness; • Had a diagnosis of dementia; • Could not understand English or were non-literate; • Provided care to a person who met the participant exclusion criteria.
Life partners in PhD studies		• Dyads not in an intimate or spousal relationship; • One partner did not have a diagnosis of PD-MCI, PDD or DLB; • Did not live together.

4.2.2.3 Recruitment sites

Participants were recruited from seven sites in England:

- Greater Manchester Mental Health NHS Foundation Trust (GMMH; original site);
- Pennine Acute Hospitals NHS Trust (PAT; original site);
- Salford Royal NHS Foundation Trust (SRFT; original site);
- University Hospital of South Manchester NHS Foundation Trust (UHSM; original site; renamed as Manchester University NHS Foundation Trust on 01/10/2017);
- Derbyshire Healthcare NHS Foundation Trust (joined in January 2017);
- North East London Foundation Trust (NELFT) (joined in January 2017), and
- North West Boroughs Healthcare NHS Foundation Trust (NWBH) (joined in April 2017).

4.2.2.4 Recruitment strategy

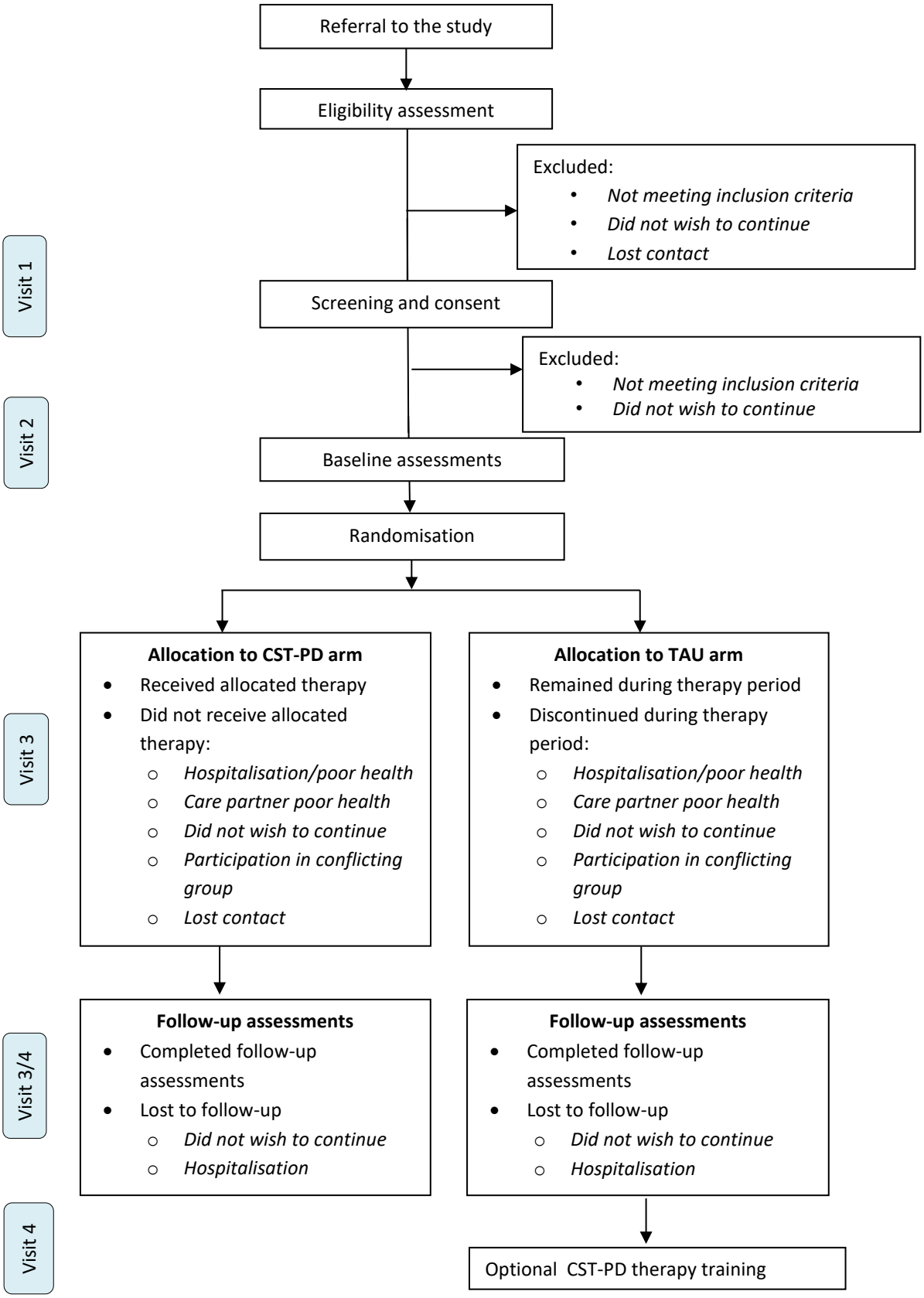
In each recruitment site the clinicians (i.e. old age psychiatrists, consultants and specialist Parkinson's disease nurses working in movement disorder clinics and memory assessment services) were informed about the INVEST study's inclusion and exclusion criteria. Potential participants with a diagnosis of PD-MCI, PDD or DLB were referred to the study team for further screening. The researchers from the NIHR Clinical Research Network team assisted with recruitment, screened participants in NHS databases and referred potential participants to me for further screening. Participants were also recruited via advertisements on the Parkinson's UK (www.parkinsons.org.uk), Lewy Body Society (www.lewybody.org) and Join Dementia Research (JDR; www.joindementiaresearch.nihr.ac.uk) websites and the study was advertised in various newsletters. Advertisement posters, leaflets about the study and INVEST newsletters (approved by the Ethics Committee, see Appendix C) were distributed at movement disorder and memory clinics across the sites with a brief

overview of the study and my contact details. A website dedicated to the INVEST study was set up to inform academics and clinicians about the study and its progress.

4.2.3 Procedure

All eligible participants were approached using best practice. If dyads were interested in the study, they received participant and care partner information sheets (Appendix C), accompanied by a cover letter, study leaflet and a sample therapy topic (Appendix C). Participant-dyads were given enough time to make an informed decision about their participation. A follow-up call with the dyad determined whether the dyad was interested and eligible to participate. If dyads were ineligible, were not interested or refused to take part they were excluded from further study-related procedures. The CONSORT flow diagram below (Figure 4.1) outlines the study procedures and visits.

Figure 4.1 Participant flow in the INVEST pilot RCT (CONSORT diagram).



4.2.3.1 Screening visit and informed consent

During the screening visit, I provided participants with clear details what the study would entail and highlighted that their participation in the study was voluntary and they were free to withdraw at any time without giving a reason. If both members of the dyad agreed to take part, they provided written informed consent. It was anticipated that participants with PRD would have capacity and be able to provide informed consent to take part in the study (MRC Ethics Guide, 2007). Separate written informed consent was sought from each member of the dyad and the dyad received a copy of the informed consent. However, if a person with PRD did not wish to take part in the study, neither member of the dyad proceeded with informed consent and enrolment to the study. Following the informed consent, I informed the general practitioners of people with PRD via letter (see Appendix C) about enrolment to the research study, unless the participant refused.

On occasions where the person with PRD had capacity to consent but the ability to write had deteriorated due to PD symptoms, the care partner wrote the participant's initials, name and date on the consent form on the participant's behalf and the participant only wrote a signature. If the participant could not write or sign the consent form, the care partner signed the participant's form with his/her own signature and I provided a written explanation that the participant had capacity to consent to the study but could not sign the form due to motor symptoms of PD.

4.2.3.2 Adults lacking capacity

An evaluation of capacity was applied according to the MRC's Ethics Guide (2007) and the Mental Capacity Act 2005. As the pilot RCT took place over 12 weeks, continuous verbal consent was taken by researchers to ensure both members of the dyad were willing to carry on participating in the study. If the person with PRD became uncomfortable during any of the researchers' visits or during the study, participation was discontinued.

On the occasions when a person with PRD lacked capacity at study entry, or lost capacity after the initial informed consent, the requirements of the Mental Capacity Act 2005 were followed and the consultee process was activated. The consultee was asked to provide an opinion on the views on whether the participant would decline to participate in the research study if he or she were to have capacity (MRC Ethics Guide, 2007). Lacking capacity means "a person is unable to make a decision for themselves because of an impairment or a disturbance in the functioning of their mind or brain" (MRC Ethics Guide, 2007, p. 9; Mental Capacity Act 2005). Assessing whether somebody has mental capacity to consent is described as a two-stage process (MRC Ethics Guide, 2007; Mental Capacity Act 2005): first, the person in question has impaired functioning of their mind or brain, and second, this impairment makes the person unable to decide whether to participate in this particular research.

The person was considered to lack capacity to decide whether to participate in a research study if they could not (based on Mental Capacity Act's 2005 Code of Practice, p. 2; MRC Ethics Guide, 2007, p.11):

- **Understand** the information related to the decision (clear and appropriate information should be provided, which might include use of simplified information sheets, images or sign language);
- **Retain** the information provided for the duration of decision-making (sufficient time should be given in order to make the decision);
- **Use** or **weigh** that information in the decision-making process (understand the consequences of each choice and of indecision);
- **Communicate** their decision (by talking, sign language or other means).

If the participant lost capacity after study entry or if the capacity fluctuated, the participant continued to take part in the study, as the initial provision of informed consent indicated agreement to participate in the study, and the agreement of a personal consultee was obtained for continuation and use of data since loss of capacity. The personal consultee is a person who has interest in the welfare of the

potential participant and who is not paid (MRC Ethics Guide, 2007). In all cases in the studies, the personal consultee was a care partner who was a member of the dyad. The role of the consultee was voluntary and they were provided with details of the study, explanation of why they had been approached and what did the role of the consultee entailed (MRC Ethics Guide, 2007).

Assessing mental capacity in every person with PRD is important as ability to make an informed choice may greatly vary and fluctuate in people with dementia (Palmer et al., 2005; Pennington, Davey, Ter Meulen, Coulthard, & Kehoe, 2018) and capacity may be diminishing already at mild cognitive impairment stage (Jefferson, Lambe, Moser, Byerly, Ozonoff, & Karlawish, 2008; Pennington et al., 2018). While many people with mild cognitive impairment and dementia are able to express their decision about whether to take part in a research study or not, they may not always be able to understand, retain and use information they were provided with to express their choice, which are cognitively more challenging tasks than expressing a decision (Jefferson et al., 2008; Pennington et al., 2018). Therefore, it was crucial to undertake an individual 'capacity to consent' assessment in each person.

Table 4.2 outlines the use of information sheets and consent forms with participants with PRD in regards to presence, fluctuation and loss of capacity. Sample copies of the Participant Information Sheets and Participant Consent Form are provided in Appendix C.

Table 4.2 The use of information sheets and consent forms with participants with PD.

Person with PRD	**Information Sheets and Consent forms**
1. Participant has capacity to consent at study entry	• Participant Information Sheet • Participant Consent Form
2. Participant lacks capacity to consent at study entry	• Participant Information Sheet • Personal Consultee Information Sheet • Personal Consultee Declaration Form
3. Participant loses capacity to consent after joining the study	• Participant Information Sheet • Consultee Post-consent continuation letter • Consultee Declaration Form for continuation

In the event that either member of the dyad wished to terminate their participation in the study, a withdrawal request form was filled (Appendix C). If the dyad was in the CST-PD arm, they no longer received or delivered therapy sessions. However, the dyads were invited to take part in the follow-up assessment, if they wished. The data that had been collected up to the point of withdrawal was used in the study analyses, unless otherwise requested by the dyad. The researcher recorded all information about participants' discontinuation in the study, including any serious adverse events (for example hospitalisation, death or other events) and reasons for loss to follow-up, which are presented in Chapter 9, Study 5, section 9.4.5.

4.2.3.3 Verifying eligibility

In order to confirm eligibility, a brief cognitive assessment, the Montreal Cognitive Assessment (MoCA, Nasreddine et al., 2005), was completed with both members of the dyad to: (1) ascertain cognitive impairment in the person with PRD, and (2) verify that the life partner was free from dementia.

The **MoCA** was originally devised to screen for mild cognitive impairment and can be administered in about 10 minutes. The MoCA assesses visuospatial abilities (clock-drawing test and three-dimensional cube copy), executive functions (alternative Trail making B task, phonemic fluency task and two-item verbal abstraction task), attention, concentration, working memory (target detection with tapping, a serial subtraction task, and digits forward and backward task), language (three-item confrontation naming task with low-familiarity animals, repetition of two syntactically complex sentences and fluency task) and orientation (questions about time and place) (Nasreddine et al., 2005). During the administration of the MoCA to the person with PRD, the tasks involving drawing (trail making B task, copying a cube, drawing a clock) and naming animals were provided as bigger drawings on a separate sheet to facilitate completion and avoid distractions of other questions.

The maximum score on the MoCA is 30 and a score of 25 or lower indicates impairment in cognition. Nasreddine and colleagues (2005) found the MoCA to be a

reliable and valid scale with high test-retest reliability and good internal consistency. Both the Parkinson Study Group (Chou et al., 2010) and the Movement Disorder Society Rating Scales Review Committee (Skorvanek et al., 2018) concluded that MoCA was the most appropriate and suitable measure to determine cognitive impairment in people with PD. The diagnostic accuracy of the MoCA for PD-MCI is 23 and for PDD 20.0-21.0 (Skorvanek et al., 2018) which was taken into account in the PhD studies.

4.2.3.4 Sociodemographic questionnaire

Following consent and eligibility assessment, a sociodemographic questionnaire was completed with the participant-dyad (see summary of questions in Table 4.3, Appendix D).

Table 4.3 Sociodemographic questions with participant-dyads.

		Person with PRD	Life partner
Socio-demographic	• Age and date of birth • Gender • Ethnicity • Educational background (highest degree and years of full-time education) • Professional background (employment/ retirement status and previous occupation) • Marital status • Living status (living with whom)	**Yes**	**Yes**
Dyadic relationship	• The relationship between the participant and the care partner (spouse or life partner) • Number of years for which the two members of the dyad have known each other	**Yes**	**Yes**
Care provision	• The duration of care provision by the life partner (in years) • The weekly care provision duration by the life partner (in hours)	**N/A**	**Yes**
Health and Parkinson's disease	• The year of Parkinson's disease diagnosis • The year when the first motor symptoms appeared and an overview of the initial motor symptoms • The year when the first cognitive symptoms appeared and an overview of the initial cognitive symptoms • Comorbidities unrelated to PD/DLB • Current prescribed medication • Timing of Parkinson's disease medication intake • Presence or absence of sensory impairment (vision, hearing) and of impulse control disorder symptoms	**Yes**	**N/A**

A motor examination with the person with PRD was undertaken after eligibility assessment and consent. The measures were:

- Movement Disorder Society Unified Parkinson's Disease Rating Scale-III motor examination subscale (UPDRS-III; Goetz et al., 2008a);
- Hoehn & Yahr disease staging (H&Y; Hoehn & Yahr, 1967); and
- Schwab and England Activities of Daily Living Scale (SE-ADL, Schwab & England, 1969).

Evaluating motor symptoms. The **UPDRS-III** (Goetz et al., 2008a) assesses the severity of various motor symptoms of parkinsonism on a five-point Likert scale, where 0 – normal, 1 – slight, 2 – mild, 3 – moderate and 4 - severe. The scale has several scores for right and left upper and lower extremities and other body distributions (e.g. face) and the maximum score for the UPDRS-III motor examination subscale is 108. A higher score on the UPDRS-III indicates a more severe symptomatology of parkinsonism. Each clinical symptom is provided with a specific description of the criteria facilitating the scoring of a symptom. The scale assesses the following motor symptoms of parkinsonism: speech, facial expression, tremor at rest, postural tremor of hands, finger taps, hand movements, leg raising ability, rigidity, arising from chair, posture, gait, postural stability and bradykinesia.

Evaluating PD stage. A five step staging system for Parkinson's disease, first introduced by Hoehn and Yahr in 1967, was used to assess the clinical stage of the person's Parkinson's disease. The **H&Y** scale ranges from Stage 1 (unilateral) to Stage 5 (debilitation and confinement). In the current work the modified H&Y scale was used for accuracy. H&Y is also undertaken with people with DLB as some people with the DLB diagnosis have been initially diagnosed with PD before being re-assessed and diagnosed with DLB.

Evaluating activities of daily living. The level of ability/disability was assessed by the **SE-ADL** Scale (Schwab & England, 1969) which has scores ranging from 100% ("*Completely independent. Able to do all chores without slowness, difficulty or*

impairment") to 0% ("*Vegetative functions such as swallowing, bladder and bowel function are not functioning; bedridden"*).

The H&Y stage was assigned by the researcher following the UPDRS-III motor examination. The SE-ADL was assigned with the help from the care partner regarding the care recipients' abilities to perform activities of daily living. The full sociodemographic assessment pack (Pack I, including MoCA, UPDRS-III, H&Y and SE-ADL scales) is provided in Appendix D.

4.2.3.5 Baseline assessment procedure

Following the screening and informed consent visit, I forwarded the dyads' details to the blinded researcher(s), who then arranged a baseline assessment visit with the dyad. Due to the nature of multiple assessments with the person with PRD and care partner, two researchers visited the dyad where possible so that the assessments could be undertaken simultaneously with both members of the dyad. Frequent rest breaks were recommended to avoid fatigue by participants. At the start of the visit the researchers asked for participants' verbal consent to continue in the study. The blinded researchers highlighted that answering the questions was voluntary and should they wished not to answer, they were given the opportunity to do so.

4.2.3.6 Randomisation

Dyads were randomised after baseline assessment visits either to the Cognitive Stimulation Therapy in Parkinson's-related dementia (CST-PD) arm or the treatment as usual (TAU) arm. Randomisation was undertaken by the Manchester Academic Health Science Centre Clinical Trials Unit, who applied a single-strata blocked randomisation, and who informed me directly about the randomisation result via telephone and confirmatory e-mail. When the randomisation was performed at the external sites, I was copied in the confirmatory e-mail to know the date of the randomisation, the researcher who randomised and the arm to which the dyads had been randomised to.

I informed participants of the randomisation result via phone-call and a postal letter (see Appendix C). All dyads had an equal chance of being allocated either to the CST-PD or the TAU arm. If dyads were allocated to the CST-PD intervention group, they received a therapy training delivered by unblinded researchers, following which they undertook the CST-PD intervention for 12 weeks, 2-3 sessions a week. If dyads were allocated to the TAU group, the dyad did not receive the CST-PD intervention. TAU was the standard treatment by the National Health Service (NHS) that the participant with PRD had been receiving so far.

4.2.3.7 Blinding

Due to the nature of the INVEST study, participants could not be blind to their allocated randomisation arm; hence, it was a single-blind study. At all Greater Manchester sites, NWBH and Derbyshire sites, one team was dedicated to undertaking screening visits, therapy training visits and weekly support phone calls (unblinded researchers), and the second team (Clinical Research Network team) acted as the blinded researchers completing both baseline and follow-up assessments with the dyad. At NELFT, all members of the research team were able to conduct baseline assessments as dyads had not been randomised at that stage and therefore the researchers could not have been unblinded. All researchers encouraged dyads not to reveal their randomisation arm to the blinded researcher during the follow-up visits. The INVEST study statistician remained blind to allocation whilst performing the main analysis for the INVEST study.

4.2.3.8 Treatment as usual (TAU)

Including a comparator group (i.e. TAU) in pilot studies is important as it permits evaluation of operational procedures such as recruitment, retention, randomisation and the implementation of the intervention (Leon, Davis & Kraemer, 2011). The purpose of including a therapy and control arm groups in the INVEST study was to compare the two groups on the primary outcome measure (cognition) for which the study was powered (McCormick et al., 2017b).

Dyads allocated to the TAU arm did not receive any supplementary intervention in addition to their standard NHS treatment for motor, psychiatric and cognitive symptoms for the person with PRD. The typical treatment for people with PRD included dopamine replacement therapy for the symptomatic relief of the PD symptoms, medication enhancing cognition (e.g. rivastigmine, memantine) and support from a PD nurse specialist and/or consultant. Due to the complex nature of PD-MCI, PDD and DLB and the individual needs of people with PRD, all participants could access specialist services such as speech and language therapy, physiotherapy, occupational therapy, psychology or psychiatry services, and any other specialist services for the specific symptoms a person with PRD was experiencing. An unblinded INVEST researcher recorded the appointments with specialists in the weekly phone-call log when becoming aware of these. If a change in medication occurred, it was recorded as an adverse event (described in section 4.2.6).

A number of participants regularly attended local voluntary sector support meetings, lunch clubs or day centres organised by Parkinson's UK, Alzheimer's Society or Age UK. Participants in both arms were allowed to continue accessing these services but it was advised that the dyad should attempt to refrain from starting participation in cognitively stimulating interventions whilst they were taking part in the INVEST study; this was monitored and recorded in the weekly phone-calls. In one occasion where a dyad initiated attendance at a reminiscence group, which shared similar aspects with the CST-PD intervention, the dyad was withdrawn from the INVEST study as this acted as a confounding factor and may have biased the final results.

4.2.4 Outcome assessments

The outcome measures were completed in an interview between the blinded researchers and the participant-dyads at baseline and follow-up. Questionnaires were divided into three parts (Appendix D): Pack IV – life partner questionnaire; Pack II – person with PRD questionnaire, and Pack III – proxy-measure completed with life partner about the person with PRD. An overview of all scales used in the PhD studies is provided below.

For the purposes of the postal questionnaire study with life partners of people with PRD, assessment Pack IV was used. Additional sociodemographic questions were included in the postal assessment such as gender, date of birth, ethnicity, education, marital status, relationship duration and living status of both members of the couple; the duration of care provision in years and hours per week by the life partner; and diagnosis, year of PD or DLB diagnosis and the year of the onset of cognitive symptoms of the person with PRD (Appendix F).

4.2.4.1 Care partner questionnaire (Pack IV)

Evaluating burden. Life partners' burden was evaluated with the **Zarit Burden Interview** (ZBI; Zarit et al., 1980), which is one of the most extensively used scales to assess life partners' physical, emotional and socio-economic status in regards to care provision. The self-report instrument consists of 22 items (for example: "*Do you feel strained when you are around your relative?"*) which are scored on a five-point Likert scale ranging from 0 (never) to 4 (nearly always). Higher scores reflect greater burden in the life partner. The ZBI has been used with life partners of people with PD (Leroi, Harbishettar et al., 2012b).

Evaluating relationship satisfaction and associated feelings of care provision. Life partners' relationship satisfaction was assessed with four scales:

(1) The **Relationship Satisfaction Scale** (RSS; Burns, 1983) evaluated the person's satisfaction with the relationship with their partner. The seven-item RSS explores the communication and openness, resolving conflicts and arguments, degree of affection and caring as well as overall satisfaction with the relationship in a seven-point Likert scale varying from 0 (very dissatisfied) to 6 (very satisfied). To my knowledge, this scale has not been in used before in PRD and the psychometric properties of the scale are discussed in Chapter 5 (Study 1).

(2) The **Dyadic Relationship Scale** (DRS; Sebern & Whitlatch, 2007) measured positive and negative aspects of the dyadic relationship within family care with an 11-item scale on a four-point Likert scale ranging from 0 – strongly disagree

to 3 – strongly agree. The DRS consists of a positive dyadic interaction subscale (items 1, 2, 6, 7, 9, 10; for example: *I felt closer to her/him that I have in a while*") and a dyadic strain sub-scale (items 3-5, 8, 11; for example: "*I felt strained*"). Higher scores indicate greater perceived strain and greater positive interaction with the person life partners care for.

(3) The **Relatives Stress Scale** (Rel.SS; Greene, Smith, Gardiner, & Timbury, 1982) assessed the amount of stress and upset experienced by the life partner as a result of having to care for the person with PRD. The Rel.SS consists of 15 items on a five-point Likert scale (0 – never/not at all, 1 – rarely/a little, 2 – sometimes/moderately, 3 – frequently/quite a lot and 4 – always/considerably) and is divided into three subscales: personal distress, life upset and negative feelings. For example, "*Do you ever feel that you need a break*" (personal distress subscale, items 1-6), "*Is your sleep interrupted by (your relative)*" (life upset subscale, items 7-11) and "*Do you ever feel frustrated at times with (your relative*)" (negative feelings subscale, items 12-15). The Rel.SS has previously been used with partners of people with PDD (Thommessen et al., 2002).

(4) **Family Caregiving Role Scale** (FCR; Schofield, Murphy, Herrman, Bloch, & Singh, 1997) was used to assess aspects related to the caregiving role with 16 items on a five-point scale ranging from 1 – strongly disagree to 5 – strongly agree. The FCR is divided into three subscales: (1) life partners' satisfaction (items 1-7), which evaluates the positive emotional response to the care recipient and the caring role (for example: "*I get a great deal of satisfaction from caring*"); (2) care partner resentment (items 8-12), which assesses negative effects on the care partners' life, opportunities, time and social relationships (for example: "*I have lost control of my life since caring for (my relative*)"; and (3) care partner anger (items 13-16), which focuses on assessing negative emotional responses to the care recipient like guilt, anger and embarrassment (for example: "*Nothing I can do seems to please (my relative)*"). To my knowledge, the FCR scale has not been used with life partners of people with PD.

Evaluating mood. Mood was evaluated with the 14-item **Hospital Anxiety and Depression Scale** (HADS; Zigmond & Snaith, 1983) where seven items measure anxiety

and seven items measure depression. Questions are worded either positively (for example: *"I feel cheerful"*) or negatively (for example *"I get sudden feelings of panic*") on a four-point Likert scale. The items vary from 0 to 3 and ask about how the person felt over the past week. Scores are divided into categories of normal (0 – 7), mild (8 - 10), moderate (11 – 14) and severe (15 – 21). The HADS is deemed to be a valid measure of anxiety and depression in care partners and in people with PD and has been frequently used in research (Marinus, Leentjens, Visser, Stiggelbout, & van Hilten, 2002; Mondolo et al., 2006; Rodriguez-Blazquez et al., 2009).

Evaluating own health. To evaluate life partners' own perceived health, the **Short Form 12 Health Survey** (SF-12; Ware, Kosinski, & Keller, 1996) was used. The SF-12 measures various limitations in role functioning as a result of physical and emotional health and consists of 12 items; four are binary (yes/no answer) and eight items are scored on a Likert response scale (Ware et al., 1996). The questions cover general health, daily activities that might have been limited by physical restraints, pain, emotional problems (for example: *During the past 4 weeks, how much did emotional problems, such as depression or anxiety, interfere with your work or other regular daily activities?)* (Ware et al., 1996). Separate sub-scores are calculated for physical and mental health.

Evaluating quality of life. Health-related quality of life was measured with the **EuroQol-5D-3L** scale (EQ-5D; The EuroQol Group, 1990), which comprises five questions regarding the person's health state today including mobility, self-care, usual activities, pain/discomfort and anxiety/depression. Each question has three answer options: no problems, some problems, or extreme problems. In addition, EQ-5D has a visual analogue scale (EQ-5D VAS) which asks the person to rate their health state today on a thermometer scale from 0-100%. EQ-5D has been used in older adults and in PD populations (Hechtner et al., 2014; Kent, Gray, Schlackow, Jenkinson, & McIntosh, 2015).

Evaluating resilience. Resilience was measured with the **Brief Resilience Scale** (BRS; Smith et al., 2008), which assesses the ability to bounce back or recover from stress.

The BRS consists of six items on a five-point Likert scale (from 1 – strongly disagree to 5 – strongly agree). Three items are worded positively (for example: "*I usually come through difficult times with little trouble*") and three items are worded negatively (for example: "*I have a hard time making it through stressful events*"). The authors of the BRS (Smith et al., 2008) have found good internal consistency and test-retest reliability.

4.2.4.2 Person with PRD questionnaire (Pack II)

Evaluating relationship satisfaction. RSS (described in section 4.2.4.1)

Evaluating quality of life. EQ-5D (described in section 4.2.4.1)

Evaluating mood. HADS (described in section 4.2.4.1)

4.2.4.3 Proxy-completed questionnaire (Pack III)

Evaluating observer-rated neuropsychiatric symptoms. The **Neuropsychiatric Inventory** (NPI; Cummings et al., 1994) was used in an interview between the life partner and the researcher regarding the presence or absence of neuropsychiatric symptoms in the person with PRD. The NPI includes the following symptoms: delusions, hallucinations, agitation/aggression, depression/dysphoria, anxiety, elation/euphoria, apathy/ indifference, disinhibition, irritability/lability and aberrant motor behaviour. The questions in the NPI refer to the changes in the care recipients' behaviour that has manifested since the onset of the disease (Cummings et al., 1994). The researcher first asked screening questions to identify whether a specific behavioural change was present or absent in the participant. If the neuropsychiatric symptom was present, the researcher proceeded with asking sub-questions about that symptom (yes-no answers). Then, the life partner was asked to provide an overall rating of frequency and severity of that particular behavioural domain. The frequency of the symptom is rated as: (1) Rarely – less than once per week, (2) Sometimes – about once a week, (3) Often – several times per week but less than every day, and (4) Very often – once or more per day. The severity is rated in a following way: (1) Mild – produces little

distress in the person, (2) Moderate – more disturbing to the person but can be redirected by the care partner, and (3) Severe – very disturbing to the person and difficult to redirect. A magnitude score for each domain is calculated by frequency x severity.

Following the ratings of frequency and severity, the life partners were asked if they felt any psychological or emotional distress due to care recipients' behaviour. If so, it was rated on a 6-point Likert scale: 0 – not at all, 1 – minimally (almost no change in work routine), 2 – mildly (some change in work routine), 3 – moderately (disrupts work routine), 4 – severely (disruptive, upsetting to other people), 5 – very severely or extremely (very disruptive, major source of distress for other people). The total NPI score is calculated by adding up the 10 domain scores (excluding the care partner distress score). The NPI is one of the gold-standard instruments for measuring neuropsychiatric symptoms in PD and has been used in PRD as well (Chiu et al., 2016).

4.2.5 Cognitive Stimulation Therapy in Parkinson's-related dementia (CST-PD)

CST-PD has been adapted from the earlier group-based CST (Spector et al., 2001; 2003) and individualised CST (Orgeta et al., 2015a; Orrell et al., 2017) for people with dementia and their study partners (described in Theory chapter section 2.1.6.3). The CST-PD intervention went through full adaptation in 2015 through separate focus groups with professionals (movement disorder consultants, nurses), people with PRD and care partners, and through individual interviews with people with PRD (McCormick et al., 2017a). The principles of the CST-PD are rooted in the CST and iCST but the intervention has been fully modified to meet the needs of people with PRD, such as excluding physical tasks due to motor issues and avoiding bright images and abstract questions due to neuropsychiatric symptoms (McCormick et al., 2017a). The full development process of the CST-PD is described elsewhere (McCormick et al., 2017a).

CST-PD is a home-based care partner-guided psychosocial intervention. The aim of the intervention is to engage in themed 20-30 minute conversations and/or activities using a therapy manual as a guide. The activities are designed to initiate thought processes,

opinions, focus, language, memory, planning and executive functioning (e.g. attention, emotion- and self-regulation), essentially stimulating the person's cognitive abilities and flexibility. Dyads are allowed to pick any topic of interest to them from the therapy manual and spend approximately 20 to 30 minutes discussing the topic. Participant-dyads are asked to complete two to three sessions a week for 12 weeks. Prior to commencing the therapy, dyads were encouraged to find photos of themselves, their family members and loved ones, and put together a memory box consisting of various meaningful objects and memorabilia (e.g. books, vinyl disks, postcards, gifts, souvenirs, etc.) to support memory, reminiscence and personalise the therapy.

4.2.5.1 Structure of CST-PD sessions

The CST-PD manual consisted of 65 topics which were divided into 9 sections (see Table 4.4). The length of each individual topic varied between 2 and 6 pages. Few sample topics are provided in Appendix E.

At the start of the session the dyad was asked to select a topic from the therapy manual. Each therapy session followed the principles of the therapy but varied in terms of content – all therapy topics included pictures and images of a particular subject area to support the memory and act as a point of reminiscence, but some topics included a game or an activity (e.g. crossword, word search, match the pairs, drawing, colouring in, etc.). As the topics progressed, the nature of the questions became slightly more difficult and cognitively demanding by thinking about 'why' and 'how'. The dyads were flexible in terms of the duration and timing of the session. Having small breaks during the therapy sessions were encouraged. The care partners completed a diary following each therapy session to record the operational aspects of the therapy session, such as date, duration, and topic title.

Table 4.4 Themes and topics from the CST-PD manual.

CST-PD session theme	Specific topics	
1. Personal Life	1.1 Childhood 1.2 My family 1.3 Relationships	1.4 Education & Occupation 1.5 Wedding traditions
2. Food	2.1 Breakfast 2.2 World cuisine 2.3 Ingredients	2.4 Then and now 2.5 Staying healthy
3. Hobbies and Leisure	3.1 My perfect day 3.2 Parks 3.3 Pets 3.4 Water sports 3.5 Ball games 3.6 Winter sports	3.7 Flowers and trees 3.8 Vegetables and herbs 3.9 In and around the garden 3.10 Libraries and reading 3.11 Space and planets
4. Art	4.1 History of art 4.2 Lowry & Turner 4.3 19th century 4.4 Pop art	4.5 Art from the Islamic world 4.6 Architecture 4.7 Drawing cartoons 4.8 Painting water
5. Media and Entertainment	5.1 Art inspired by music 5.2 Musical instruments 5.3 Music genres 5.4 Live performances	5.5 Current affairs: Magazines 5.6 Current affairs: Newspapers 5.7 Current affairs: Reporters 5.8 Technology
6. Nature	6.1 Patterns, shapes and colours 6.2 Cloud formations 6.3 Weather conditions	6.4 Water 6.5 Water and people 6.6 Animal kingdom
7. Seasons	7.1 Autumn 7.2 Winter	7.3 Spring 7.4 Summer
8. Travel and Culture	8.1 Continents 8.2 UNESCO sites in Europe 8.3 Flags 8.4 Seven Human Wonders 8.5 Public celebrations	8.6 World celebrations 8.7 Chinese New Year 8.8 Blackpool: Illuminations 8.9 Blackpool: Performances 8.10 Blackpool: The holiday destination
9. Games	9.1 Old Wives' tales 9.2 Being creative 9.3 Tic tac toe 9.4 Match the pairs	9.5 Decode the sentence 9.6 Proverbs 9.7 Quiz board game 9.8 Colouring and doodling

4.2.5.2 Principles of CST-PD

The dyads were asked to follow the 9 key therapy principles of CST-PD (See Table 4.5) which are derived and adapted from the CST and iCST interventions.

Table 4.5 CST-PD principles with descriptions.

Therapy principle	Brief overview
1. Consider the person's needs	Focus on the person rather than memory problems and impairment. Incorporate the person's interests in the session and tailor it according to their needs.
2. Offer choice	Encourage the person to choose a topic or select a topic together at random. Ascertain that the chosen topic will be engaging and at the right difficulty level.
3. Focus on opinions rather than facts	Ask opinion-based questions, rather than fact-based questions, as there are no right or wrong answers. In case of fact-based elements in the activities, provide a selection of options or cues (e.g. images) to facilitate finding an answer.
4. Have a tangible focus	Incorporate and combine senses (vision, hearing, touch, taste and smell) which can stimulate memory. You can put together a memory box which helps with concentration, memory and personalising the therapy.
5. Use reminiscence	Support the person by helping them to reminisce about their past memories and experiences and be sensitive when discussing sad or negative memories.
6. Maximise potential	Explore the person's potential (e.g. 'comfort zone') to know their capabilities and provide enough prompting so that they can carry out the activities but be flexible, patient and allow plenty of time.
7. Enjoyable and fun	Engage in the therapy in a fun and enjoyable way so that the person can feel comfortable, enabled and empowered.
8. Stimulate new ideas and communication	Provide the person with additional questions to encourage discussion, new ideas, thoughts and associations, rather than solely focusing on recalling previously learned information. You may wish to use your own ideas and resources.
9. Strengthen the caregiving relationship	It is important to assure that you have both rested before the session and made time in order to enjoy the therapy together at your own pace and time. Alternatively, you may pick another date and time to complete the session.

4.2.5.3 CST-PD training

The dyads allocated to the CST-PD arm were trained by myself or other unblinded researchers following baseline assessments. During the training, the dyads received a CST-PD manual, care partner diary (see example sheet in Appendix E), training materials with the background to the intervention, 9 key principles (described in Section 4.2.5.2) and role play (see Appendix E). At the start of the training the researcher introduced the intervention, explained the 9 key therapy principles and completed a role play whereby the researcher was in the role of the care partner and the care partner was in the role of the person with PRD receiving the intervention. The purpose of the role-play was to demonstrate the therapy to the care partner so that they could experience the therapy as a recipient.

Then, the researcher proceeded to complete a session with the person with PRD whilst the care partner observed. This was an example therapy session to demonstrate some of the key principles. After a few minutes the researcher encouraged the care partner to join in and lead the therapy session and the researcher provided constructive feedback on their proficiency in guiding the therapy session and applying the therapy principles. This was done to improve care partners' skills and confidence and raise awareness of their adherence to the training protocol.

After training the CST-PD arm dyads and informing the TAU arm dyads about their allocation, all dyads entered into a two-week lead-in period. The purpose for the CST-PD arm was to allow familiarisation with the therapy and decide whether they wished to continue with the 10-week intervention or not, and for the TAU arm to allow a similar duration of participation in the study (approximately 12 weeks). After two weeks, the unblinded researcher queried the CST-PD arm dyads about their experience with the therapy, the number of therapy sessions completed, and whether they wished to proceed with the 10-week intervention, and provided the dyad with support and guidance on the therapy, if they needed it. If the therapy arm dyad was happy to receive the intervention, they could start with the 10-week CST-PD. At this time, the control arm participants could also start their 10-week TAU. All dyads were informed

that they would start receiving weekly phone calls from the research team from the next week onwards.

All dyads received weekly phone calls from myself or other unblinded researchers. It was deemed important that participants in the CST-PD and TAU arms received the same amount of contact from the researcher to minimise the drop-out rates of participants. The researcher recorded the date of the phone-call (if contact was made), any serious adverse events or adverse events (see section 4.2.6), and planned holidays, and asked the CST-PD arm dyads for the number and duration of sessions in the past week and whether they required additional support and wrote a brief summary of the dyads' progress with CST-PD.

<u>The telephone assistance ensured the following:</u>

a) Verbal consent in continuing taking part;
b) Recording of any (serious) adverse events since past week;
c) Recording any planned holidays during the study;
d) Confirming that the participant was not taking part in any other dementia-related intervention research study;
e) Ensuring sufficient differentiation between the two arms;
f) *Number and duration of the therapy sessions in the past week;
g) *The regular completion of the therapy diaries by care partner to record the CST-PD sessions;
h) *The confidence of care partners in delivering the therapy and in managing any challenging situations that may have occurred during the therapy sessions;
i) *Necessity of receiving optional booster therapy training session, if requested by the care partner or if the need was identified by the researcher.

** CST-PD arm only*

4.2.5.4 Following 12-weeks of intervention

After 12-week participation in the study, the blinded researchers carried out follow-up data collection visits with all dyad. The outcome results for life partners in the CST-PD and TAU arms are described in Chapter 9 (Study 5).

The dyads that were initially allocated to the control (TAU) group were invited to take part in the second phase of the 12-week CST-PD intervention (cross-over arm) as a courtesy, provided that they had completed the follow-up assessments. The therapy training for the cross-over arm dyads was delivered according to the protocol (described in section 4.2.5.3) but they did not undertake any further study-related procedures or assessments and had therefore formally exited the study. However, they were encouraged to contact the research team if they needed support or assistance with delivering the therapy.

4.2.6 Safety reporting

All researchers recorded any adverse events (AE) and serious adverse events (SAE) on becoming aware. An SAE was considered to be an untoward event experienced by a participant or care partner which (a) was life-threatening, (b) resulted in death, (c) required hospitalisation or prolongation of existing hospitalisation, (d) resulted in persistent or significant disability or incapacity, or (e) was otherwise considered medically significant by the chief investigator (see Appendix C). An AE included incidents, protocol violations and protocol deviations (see Appendix C). CST has not been found to be associated with adverse events, therefore it was anticipated that adverse events would not occur due to CST-PD intervention. In the rare instances when an AE or SAE occurred, the dyad was directed to relevant health services, and the event was recorded on a specific AE or SAE form. Following the event, advice was sought from the Chief Investigator (I.L.) in regards to further action and whether a withdrawal of the dyad should be considered. The dyad was informed of the decision made by the Chief Investigator.

4.2.7 Analyses

All quantitative data were analysed in IBM Statistical Package for Social Sciences software for Windows Version 23.0 (SPSS, IBM Corp, 2015) using descriptive and inferential statistics, as appropriate. Categorical variables were presented as percentages and continuous variables as means and standard deviations (normally distributed data) or medians and interquartile ranges (non-normally distributed data). The data were examined for any missing values with Little's Missing Completely at Random (MCAR; Little, 1988) and missing data were addressed in each study, as appropriate, to ensure integrity of the data set. Assumption tests were undertaken in each study to verify the appropriateness of the parametric analyses. Depending on the analyses, the following assumption tests were included: normality of residuals, linearity, outliers, homoscedasticity, independence of observations and/or residuals, lack of multicollinearity, homogeneity of variance, or homogeneity of regression slopes. If one or more of the assumption tests were not met, the non-parametric analyses were used. Post hoc comparisons were applied in studies where it was deemed important. The threshold for significance was set at $p < 0.05$ and confidence intervals were recorded where deemed appropriate.

A number of analyses were undertaken which are summarised below and detailed in individual study chapters:

- Descriptive statistics (Studies 1-5)
- Internal reliability (Cronbach's α) (Studies 1 and 2)
- Psychometric properties: convergent validity, floor and ceiling effects (Study 1)
- Correlation analyses (Pearson, Spearman) (Studies 1, 2, and 4)
- T-test (Studies 1 and 4)
- ANOVA (Studies 1 and 2)
- Exploratory factor analysis (Study 2)
- Regression analysis (Study 2)
- Thematic analysis (Study 3)
- Multilevel modelling (Study 4)
- ANCOVA (Study 5)

4.2.7.1 Data entry

All data from the baseline and follow-up questionnaires were entered throughout the duration of the study. I was responsible for receiving the data from all participating sites. The data were entered item-by-item by an independent data entry clerk into SPSS datasets, which were used in the quantitative studies. The data for each participant and care partner was entered on a separate row (one row per person), and linked via dyad ID (e.g. D001).

Upon completion of the data entry by the data entry clerk, I checked 100% of the data entries to ascertain maximum accuracy of data entry. The study co-ordinator (S.A.M.) then verified the correctness of the data by inspecting 10% of the baseline and follow-up data entries. Once the datasets were populated, I looked through the datasets for inconsistencies and missing data. I fully entered the data from the postal questionnaire survey and this did not undergo further data entry checks.

4.2.8 Ethical considerations

The INVEST study received a favourable opinion by the NHS National Research Ethics Service (NRES) Yorkshire & the Humber – Bradford Leeds Research Ethics Committee (15/YH/0531; See Appendix A) in January 2016. The study followed the guidelines of the Declaration of Helsinki and the guidelines of Good Clinical Practice. All researchers involved in the study received Good Clinical Practice training. The protocol and the study documents were reviewed by the Research and Development departments in all participating sites and the University of Manchester.

The documentation of the postal questionnaire study with life partners of people with PRD, was submitted as a Substantial Amendment to the NRES Yorkshire & the Humber – Bradford Leeds Ethics Committee and received a favourable opinion in June 2017. The documents that were submitted included a participant information sheet, consent form, invitation letter and the questionnaire (see Appendix F).

4.2.8.1 Confidentiality and data management

The research was undertaken in full compliance with the Data Protection Act 1998 and researchers were responsible for preserving the confidentiality of the data at all times. All participants were reassured that their data were anonymised and kept confidential, and that no personal information would be identifiable nor published in any research studies. Blinded researchers who carried out baseline and follow-up assessments with dyads transferred the data to the research team in a timely and secure manner. Personal information (such as name, contact details, date of birth) of participants was stored separately from the data.

Participants were assigned a unique individual identification code and a dyad code, which was used in all data storage files to anonymise any written or electronic document and any audio or data set file. Names or other personal identifiable information were not used in the code. The audio files, which were recorded with digital voice recorders, were saved onto an encrypted computer and deleted from the digital voice recorders. The interview transcripts were fully anonymised and password protected. All research documents will be kept securely for fifteen years at the sponsor's site after study termination.

CHAPTER 5: The profile of life partners of people with Parkinson's-related dementia: Does clinical syndrome matter? (Study 1)

This chapter describes a cross-sectional observational study of life partners of people with Parkinson's-related dementia (PRD) and is currently under review by the Journal of Geriatric Psychiatry and Neurology.

5.1 Introduction

Parkinson's disease (PD) is a complex movement disorder encompassing motor, psychiatric and cognitive symptoms. About a quarter of people who have been newly diagnosed with PD present with mild cognitive impairment (PD-MCI) (Aarsland et al., 2009, 2010; Foltynie et al., 2004; Muslimovic et al., 2005) and between 15% and 57% develop PD-MCI following the PD diagnosis (Yarnall, Rochester, & Burn, 2013). Once cognitive impairment has emerged in PD, the likelihood of progressing to PD dementia (PDD) increases considerably (Goldman et al., 2018a). It is estimated that approximately 80% of people with PD develop dementia within 10 to 20 years following the diagnosis of PD (Aarsland et al., 2003; Hely et al., 2008). This is important as the prevalence of neurodegenerative conditions such as PD and PDD are an increasing trend due to the aging population and the prevalence of PD is expected to rise twofold and the prevalence of PDD threefold by 2060 (Savica et al., 2018). Another common type of dementia, which shares underlying pathology and cognitive and neuropsychiatric manifestations with PDD, is dementia with Lewy bodies (DLB), which has a prevalence of 4.2-4.6% of all dementia cases (Kane et al., 2018; Vann Jones & O'Brien, 2014). Collectively, PD-MCI, PDD and DLB belong under the umbrella term of 'Lewy body spectrum disorders' (Aarsland, 2016; Goldman et al., 2014), but in the current chapter the term 'Parkinson's-related dementia' (PRD) will be used throughout.

The progressive and demanding nature of PRD necessitates the support of a care partner, a role which is frequently fulfilled by spouses, life partners, adult children or

other family members (Ham, 1999; Pearlin et al., 1990). Care partners play a crucial role in supporting disease management and activities of daily living of people with PRD. However, providing care may lead to challenges in balancing personal and care-related responsibilities, which can in turn increase burden and stress (Carter et al., 2008; Leiknes et al., 2015; Lökk, 2008; Martinez-Martin et al., 2008, 2015; Mosley et al., 2017; Whetten-Goldstein et al., 1997) and contribute to increased levels of depression and anxiety (Martinez-Martin et al., 2008; Schrag et al., 2006). With the progression of cognitive impairment in PD, the *care burden* (Cifu et al., 2006; Jones et al., 2017; Leroi et al., 2012a; Martinez-Martin et al., 2015; Thommessen et al., 2002) and *emotional stress* (Lawson et al., 2018) intensifies in care partners, whereas care partner *quality of life* drops (Leroi et al., 2012a). These findings suggest that the stage of cognitive impairment in the person with PD has a significant negative impact on care partner outcomes.

In the UK, there are currently 6,5 million care partners of whom 11% provide care to a person with dementia (Carers Trust, 2015). The financial contributions by care partners thanks to the care they provide to care recipients exceeds that of the annual budget of the NHS in England, totalling to about £132 billion per year (Carers UK, 2018). The role and profile of care partners of people with PD and non-PD type dementia has been described well already. Typically, a care partner of a person with PD is a 70-year-old female spouse, who lives with the care recipient and has provided care for an average of 5 years (Cifu et al., 2006; Hand et al., 2018; Lökk, 2008; Peters et al., 2011). In Hand and colleagues' study (2018), care partners of people with moderate to late stage PD provide up to 16 hours of care each day helping with housework, assisting with personal care, being there for their partners as a friend and a companion as well as ensuring safety of their partner. Importantly, care partners are often older adults themselves and nearly half have physical or mental health problems (Hand et al., 2018), which makes caring for one's partner increasingly demanding whilst also taking care of oneself.

The heightened focus by care partners on caring for people with PRD may lead to neglect of care partners' own health and needs (Birgersson & Edberg, 2004; Pinquart &

Sörensen, 2003a) and even burnout (Mosley et al., 2017). As a consequence, some care partners may no longer be able to cope with the situation and at this stage people with PRD may likely be admitted to residential and nursing care homes, which significantly raises health and social care costs (Boström, Jönsson, Minthon, & Londos, 2007; Low, Ben-Shlomo, Coward, Fletcher, Walker, & Clarke, 2015; Mueller et al., 2017; Rockwood, Stolee, & McDowell, 1996; Tison et al., 1995; Vossius et al., 2014). Thus, it is important to understand the profile of care partners in order to provide targeted and efficient interventions for care partners.

Several studies have compared outcomes of care partners of people with varying stages of cognitive impairment in PD, typically PD with no cognitive impairment, PD-MCI and PDD (i.e. Lawson et al., 2017; Leroi et al., 2012a); however, there is little understanding of the profile of caregiving spouses or life partners in the context of PD-MCI, PDD and DLB. Thus, the current study aimed to examine the profile of caregiving life partners of people with PRD and provide comparisons of life partner outcomes according to the stage of cognitive impairment in PD. An additional objective of the study was to assess the psychometric properties of the outcome measures of life partners. A number of scales have been frequently used to evaluate outcomes in care partners of people with PRD but most studies do not report on the psychometric properties of these scales and knowledge regarding recommended scales in this population is currently lacking. Many researchers, academics and clinicians may face challenges in choosing the most suitable and appropriate instrument which has robust psychometric properties and is short, informative, easy to administer and does not add to the already existing burden of care partners (Elf, Nordin, Wijk, & Mckee, 2017; Hudson et al., 2010). Therefore, gaining insight into what measures are suitable, valid, reliable and usable is important in order to compare, contrast and standardise evidence regarding life partners' outcomes in the context of PRD, and helps researchers with the decision-making regarding scale selection for future studies.

5.2 Aim

The objectives of Study 1 were threefold: (1) to describe the sociodemographic and clinical syndrome of life partners of people with PRD, including physical and mental health, burden, stress, quality of life and feelings related to care provision, (2) to compare life partners' outcomes according to the clinical syndrome (PD-MCI, PDD or DLB), and (3) to evaluate psychometric properties of the scales to provide recommendations for future studies.

5.3 Methods

Chapter 4 provides a detailed overview of the design, eligibility criteria, recruitment, procedure, data collection, outcome measures and ethics of Study 1. The following sections will briefly summarise the methods and analyses relevant to the current study.

5.3.1 Research design

This was a cross-sectional questionnaire-based study with life partners of people with PD-MCI, PDD and DLB. Full ethical approval for the study was granted by the Yorkshire & the Humber – Bradford Leeds Research Ethics Committee (reference number: 15/YH/0531) (see Chapter 4, Section 4.2.8 and Appendix A). Informed written consent was obtained from all participants prior to participation and all participants had the capacity to consent to the study.

5.3.2 Participant sample and recruitment

Life partners of people with PRD were recruited through two different routes: (1) a face-to-face home-based interview conducted by a researcher (undertaken as part of the baseline assessment visit in the INVEST study which ran between April 2016 and July 2017), and (2) a UK-wide postal questionnaire study (which ran between July 2017 and January 2018). Participants were eligible to take part in the study if they were a

partner or a spouse of a person diagnosed with PD-MCI, PDD or DLB, and if they lived together. Participation of people with PRD was not required in this study; however, participants who took part in the postal questionnaire study provided information about people with PRD.

In route two, spouses and life partners of people with PRD were identified for the postal questionnaire through: (1) a list of potential life partners in the INVEST study who screen-failed but were eligible for the postal questionnaire, (2) patient databases at the Greater Manchester Mental Health (GMMH) and North West Boroughs Healthcare (NWBH) NHS Trusts, and (3) advertisements on Parkinson's UK, Lewy Body Society and Join Dementia Research websites.

5.3.3 Procedure

Following ethical approval in June 2017 to recruit a sub-sample of life partners, the postal questionnaire packs were prepared for posting out to potential participants. Each questionnaire pack contained an invitation letter, a participant information sheet, a consent form, a survey and a pre-paid envelope with my postal address (see Appendix F). My full contact details were provided in case participants wished to find out more about the study prior to taking part, receive help in completing the questionnaire or ask any questions related to the study.

First, the questionnaires were posted out to spouses and life partners who had screen-failed participation in the INVEST study due to ineligibility, distance, high presence of burden in the life partner or lack of interest in participating. Second, colleagues at GMMH and NWBH NHS Trusts identified potential participants via patient databases and sent the postal questionnaires out to the partners of people with PRD. Third, an advertisement together with a brief introduction of the study and a participant information sheet was available on the Parkinson's UK website alongside with my contact details. The study was also advertised in monthly research-related Parkinson's UK newsletters. Participants who were interested in taking part in the study contacted me via e-mail or telephone and I verified whether they were eligible to participate

before posting out a questionnaire. Fourth, the study was advertised on the Lewy Body Society's website. Fifth, a brief description of the study and its aims were available on the Join Dementia Research website.

Once questionnaires were posted to potential respondents the participants were asked to read an invitation letter and a participant information sheet and decide whether they were happy to take part. If life partners agreed to participate, they were asked to complete a consent form and a questionnaire. If potential participants decided not to take part, they were asked to dispose of the questionnaire pack and the survey. Following the completion of the questionnaire, participants returned the consent form and the questionnaire by post to me. Ethical approval to follow up participants with a telephone call, if they had not returned the questionnaire, was not sought.

5.3.4 Measures

The life partners completed a battery of socio-demographic questions (i.e. age, gender, education, ethnicity), details about partners' disease (e.g. diagnosis, disease duration) and care provision (weekly care provision hours, years of care provision) as well as the following measures (described in detail in Chapter 4, section 4.2.4.1):

- The Relationship Satisfaction Scale (RSS; Burns, 1983),
- The Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983),
- The Zarit Burden Interview (ZBI; Zarit et al., 1980),
- The Short Form 12 Health Survey (SF-12; Ware et al., 1996),
- The EuroQoL-5D-3L (EQ-5D; The EuroQol group, 1990),
- The Relatives' Stress Scale (Rel.SS; Greene et al., 1982),
- The Dyadic Relationship Scale (DRS; Sebern & Whitlatch, 2007),
- The Family Caregiving Role Scale (FCR; Schofield et al., 1997),
- The Brief Resilience Scale (BRS; Smith et al., 2008).

Furthermore, specific disease symptoms of people with PRD could be elicited from the home-based assessments, including (see Chapter 4, section 4.2.3.4):

- cognition (measured with the Montreal Cognitive Assessment, MoCA, Nasreddine et al., 2005),
- severity of PD (measured with the Hoehn & Yahr stage, H&Y, Hoehn & Yahr, 1967),
- motor symptoms (measured with the Unified Parkinson's Disease Rating Scale part III, UPDRS-III, Goetz et al., 2008a),
- functional ability (measured with the Schwab & England Activities of Daily Living scale, SE-ADL, Schwab & England, 1969), and
- neuropsychiatric symptoms (measured with the Neuropsychiatric Inventory, NPI, Cummings et al., 1994).

5.3.5 Analyses

All statistical analyses were undertaken in IBM Statistical Package for Social Sciences software for Windows, version 23.0 (SPSS, IBM Corp, 2015). Descriptive statistics for *categorical variables* are presented as percentages, whereas for normally distributed *continuous variables* as means and standard deviations and for non-normally distributed *continuous variables* as medians and interquartile ranges. Prior to deciding which inferential statistics to use for correlation analyses and group difference analyses, data were examined for normality of distribution with the Shapiro-Wilk's test and with visual inspection of the histograms. In addition, the assumptions of linearity, outliers, homoscedasticity, independence of observations, and homogeneity of variance were evaluated to verify the appropriateness of parametric or non-parametric tests. If these assumptions were met, parametric tests were used (i.e. Pearson correlation; independent samples t-test; analysis of variance, ANOVA). However, if any of the assumptions were not met, the corresponding non-parametric tests were used as required (i.e. Spearman correlation coefficient, Mann-Whitney U-test and Kruskal-Wallis H test). Post hoc tests (i.e. Bonferroni, Hochberg or Games-Howell, as

appropriate) were used for multiple comparisons due to the use of several tests and several groups. The significance level for the results was set at $p < 0.05$.

To decide how to address the missing values in the dataset, Little's Missing Completely at Random (MCAR; Little, 1988) test was carried out in SPSS. The Little's MCAR chi-square test result [(2539, N = 136) = 2293.203, p = 1.000] revealed that the data were missing at random, meeting the assumption for undertaking an imputation method (e.g. expectation-maximization) to populate missing values. In the current study some missing information occurred in 1% of total values, in 58.0% of all the variables and in 35 participants from a sample of 136. On a variable level, missing data occurred between 0.7% and 6.7% (between 1 and 9 missing values per variable, respectively). There was a monotone pattern of missingness as assessed in the visual inspection of the missing value patterns matrix (Appendix G). Any missing data that occurred were imputed with the expectation-maximization method.

As many of the scales had not been used with life partners of people with PRD, it was important to assess the psychometric properties of these scales. Therefore, internal reliability (Cronbach α) and convergent (correlation) validity, alongside with descriptive statistics, floor and ceiling effects, and completion rate percentage are presented in the results section. The convergent validity looks at whether similar tests that are expected to be related are in fact related. The floor and ceiling effects were significant if more than 15% of participants achieved the highest or lowest possible numeric score. Calculating the floor and ceiling effects is important because high floor and ceiling effects could pose a challenge in differentiating participants from one another (Terwee et al., 2007; Wamper, Sierevelt, Poolman, Bhandari, & Haverkamp, 2010).

5.4 Results

5.4.1 Participants

A total of 136 life partners of people with PRD took part in the study. In route one, 57 life partners, who met the eligibility criteria for the current study, were included from

the INVEST study. In route two, a total of 186 postal questionnaires were posted out and 80 life partners returned the questionnaire (response rate 43%); however, one participant was excluded due to the care recipient living in a care home.

5.4.2 Characteristics of life partners and people with PRD

The characteristics of participants and their partners with PRD are provided in Table 5.1. All couples lived together and 94.9% of life partners were married. The median relationship duration was 46.5 years (IQR = 34.75, 53.00; range 5-68 years). The majority of participants were women (85.3%), white British (89.7%) with a mean age of 69.44 years (SD = 7.62; range 48-85 years). In terms of education, 61.8% of life partners had a further education qualification or a university degree but the median age of leaving full-time education was 17 years (IQR = 16 to 20; range 14 to 53). Participants had provided care for between 0 and 20 years (median = 4; IQR = 2 to 7) and were currently providing care between 0 and 168 hours per week (median = 84; IQR = 38.5 to 168). Nearly half of the participants (46.0%) provided over 100 hours of care per week.

The people with PRD were mostly male (85.3%) and white British (90.4%), with a mean age of 73.51 (SD = 6.48; range 49-90). Over half (55.9%) of people with PRD had completed further education or higher education. Thirty-seven people had a diagnosis of PD-MCI, 50 of PDD and 49 of DLB. The median disease duration of PD (including DLB diagnosis in the absence of PD diagnosis) was 5 years (IQR = 3 to 10; range 0-37) and of cognitive impairment 4 years (IQR = 2 to 6; range 0.2-22). From the sub-sample of people with PRD, who were recruited in route one, half (49.1%) had an H&Y stage of 2.0.

5.4.3 Profile of life partners

Regarding mental health, 35 life partners (25.7%) experienced clinically significant anxiety and 16 life partners (11.8%) clinically significant depression according to the HADS (see Table 5.2). The scores of the Relatives' Stress scale and the Zarit Burden

Interview showed that 81 (59.6%) and 49 (36.0%) of participants experienced stress and burden, respectively; however, 106 participants (77.9%) reported competent ability to bounce back from a negative experience (measured by the Brief Resilience Scale). A large proportion of participants (n = 79, 59.1%) were dissatisfied with the relationship as determined by the Relationship Satisfaction Scale. About half of life partners reported relatively good quality of life according to the EQ-5D-index scores (n = 67, 49.3%) and visual analogue scale (n = 71, 52.2%). Overall, the majority of participants reported satisfaction with their caring role (n = 132, 97.1%); however, over 60% of life partners displayed resentment (n = 85, 62.5%) and over 30% anger (n = 43, 31.6%) due to their caring role (measured with the Family Caregiving Role scale).

Table 5.1 Participant characteristics (n = 136 life partners).

	Life partners	People with PRD
All participants (n = 136)		
Categorical variables, N (%)		
Gender, female	116 (85.3)	20 (14.7)
Ethnicity, white British	122 (89.7)	123 (90.4)
Relationship status		
Married	129 (94.9)	
Cohabiting	7 (5.1)	
Living status		
With spouse/partner	134 (98.5)	
With spouse/partner + other family	2 (1.5)	
Education		
Left school aged 14-16 years	41 (30.1)	54 (39.7)
Left school aged 17-18 years	11 (8.1)	6 (4.4)
Further education	34 (25.0)	36 (26.5)
Higher education (university degree)	50 (36.8)	40 (29.4)
Diagnosis		
PD-MCI		37 (27.2)
PDD		50 (36.8)
DLB		49 (36.0)
Continuous variables, Mean (SD); range		
Age, years	69.44 (7.62); 48-85	73.51 (6.48); 49-90
Continuous variables, Median (IQR); range		
Relationship duration, years	46.5 (34.75, 53.00); 5-68	
Age left full-time education	17 (16.00,20.00); 14-53	16 (15.00, 20.75); 14-46
Duration of PD, years		5 (3, 10); 0-37
Duration of cognitive impairment, years		4 (2, 6); 0.2-22
Care provision duration, years	4 (2.00, 7.75); 0-20	
Care provision hours/week	84 (38.5, 168); 0-168	
Route 1 sub-sample (n = 57), N (%)		
Categorical variables, N (%)		
Retired, 'yes'	47 (82.5)	57 (100)
H&Y stage		
I		9 (15.8)
II		28 (49.1)
III		6 (10.5)
IV		10 (17.5)
V		2 (3.5)
Continuous variables, Median (IQR); range		
MoCA score (max 30)	28 (26, 30); 22-30	19 (15, 19); 7-30
UPDRS-III (max 100)		32 (30, 40); 10-58
SE-ADL (max 100)		55 (30, 80); 10-90

Abbreviations: DLB – Dementia with Lewy bodies; H&Y – Hoehn & Yahr scale; IQR – interquartile range; MoCA – Montreal Cognitive Assessment; PD – Parkinson's disease; PDD – Parkinson's disease dementia; PD-MCI – Parkinson's disease and mild cognitive impairment; PRD – Parkinson's-related dementia; SD – standard deviation; SE-ADL – Schwab & England Activities of Daily Living scale; UPDRS-III – Unified Parkinson's disease Rating Scale part III.

Table 5.2 Descriptive values of life partner outcomes (n = 136 life partners).

Description			Results								
Scales *(mode of scoring, number of items)*	**Range of scores**	**Cut-off score**	**Mean**	**SD**	**Median**	**IQR**	**Comple-tion rate N (%)**	**Actual item range**	**Alpha**	**Floor/ ceiling effects %**	**Low/ high cut-off N (%)**
RSS *(sum,7)*	0-42	≤30 indicates dissatisfaction	24.97	11.94	25	15, 35	133 (97.8)	0-42	0.944	2.9/ 8.1	79 (58.1) / 54 (39.7)
ZBI *(sum,22)*	0-88	≥41moderate/ severe burden	36.83	16.31	36	24, 48	127 (93.3)	2-78	0.922	0.7/ 0.7	78 (57.4) / 49 (36.0)
BRS *(mean,6)*	1-5	≤2.99 low resilience	3.52	0.80	3.67	3.00, 4.00	136 (100)	1.50-5.00	0.888	0.7/ 2.2	30 (22.1) / 106 (77.9)
HADS-anxiety *(sum,7)*	0-21	≥11 high anxiety	7.56	4.54	7	4, 11	134 (98.5)	0-18	0.870	2.9/ 2.2	99 (72.8) / 35 (25.7)
HADS-depression *(sum,7)*	0-21	≥11 high depression	5.78	4.07	5	2, 9	134 (98.5)	0-17	0.858	4.4/ 0.7	118 (86.8) / 16 (11.8)
SF-12-PCS *(algorithm,6)*	13-69	N/K	50.38	10.80	52.22	42.13, 59.44	134 (98.5)	24.34-68.02	N/A	0.7/ 0.7	N/A
SF-12-MCS *(algorithm,6)*	10-70	N/K	44.86	10.50	45.67	38.40, 53.50	134 (98.5)	17.56-62.63	N/A	0.7/ 0.7	N/A
EQ-5D index *(algorithm,5)*	-1.000 ... 1.000	≥0.800	0.770	0.236	0.796	0.691, 1.000	136 (100)	-0.135 … 1.000	N/A	0.7/ **27.2**	69 (50.7) / 67 (49.3)
EQ-5D VAS *(single-score,1)*	0-100%	≥80	75.20	17.30	80	65, 90	134 (98.5)	10-100%	N/A	0.7/ 1.5	63 (46.3) / 71 (52.2)
Rel.SS *(sum,15)*	0-60	≥23 stress	25.74	10.83	25	17.00, 33.50	133 (97.8)	4-55	0.898	0.7/ 0.7	52 (38.2) / 81 (59.6)
DRS-interaction *(sum,6)*	0-18	N/K	9.33	3.25	9	7, 12	130 (95.6)	2-18	0.724	0.7/ 1.5	N/A
DRS-strain *(sum,5)*	0-15	N/K	5.32	3.37	6	2, 8	127 (93.3)	0-11	0.846	9.6/ 0.7	N/A
FCR-satisfaction *(mean,7)*	1.00-5.00	≥2.5	4.04	0.51	4.00	3.71, 4.43	134 (98.5)	2.14-5.00	0.661	0.7/ 2.9	2 (1.4) / 132 (97.1)
FCR-resentment *(mean,5)*	1.00-5.00	≥2.5	2.77	0.97	2.80	2.00, 3.40	136 (100)	1.00-5.00	0.804	5.9/ 0.7	51 (37.5) / 85 (62.5)
FCR-anger *(mean,4)*	1.00-5.00	≥2.5	1.98	0.83	2.00	1.25, 2.50	135 (99.3)	1.00-4.25	0.729	**24.3**/ 0.7	92 (67.7) / 43 (31.6)

Abbreviations: Alpha – Cronbach α; BRS – Brief Resilience Scale; DRS – Dyadic Relationship Scale, positive interaction or negative strain sub-scale; EQ-5D – EuroQoL index or visual analogue scale (VAS); FCR – Family Caregiving Role scale; HADS – Hospital Anxiety and Depression Scale; IQR – interquartile range; N/A – not applicable; N/K – not known; Rel.SS – Relatives' Stress Scale; RSS – Relationship Satisfaction Scale; SD – standard deviation; SF-12 – Short Form 12 Health Survey, physical health (PCS) or mental health (MCS) sub-scale; ZBI – Zarit Burden Interview.

5.4.4 Outcome analyses

5.4.4.1 Associations between variables

Before undertaking correlation analyses, t-tests and ANOVAs to observe the associations of life partner outcomes, assumption tests were undertaken which are described in detail in Appendix G. Since not all the assumptions of Pearson correlation analyses were met, the Spearman rank correlation analyses were used. However, the assumptions for the t-tests and ANOVAs that were not met were addressed (Appendix G).

Several associations were found between life partner outcomes. Tests of the 15 variables were conducted using Bonferroni adjusted alpha level of 0.003 per test (0.05/15). Spearman rank correlation analyses showed that **lower relationship satisfaction (RSS)** in life partners was associated with higher burden (ZBI), stress (Rel.SS), anxiety (HADS), depression (HADS), negative strain (DRS), feelings of resentment and anger due to care provision (FCR), and lower mental health (SF-12), quality of life (EQ-5D), less resilience (BRS) and positive interaction with one's partner (DRS) (all at $p < 0.003$ level). **Burden** (ZBI) significantly correlated with stress (Rel.SS), strain (DRS), depression (HADS), anxiety (HADS), mental health (SF-12), quality of life (EQ-5D), resilience (BRS) and feelings of resentment and anger (FCR) (all at $p < 0.003$ level). **Lower life partners' mental health** (SF-12) was related to intrapersonal aspects (i.e. own anxiety, depression, quality of life, resilience) as well as interpersonal aspects (i.e. burden, stress, strain, resentment and anger related to care provision) (all at $p < 0.003$ level). All associations between the outcome variables are provided in the correlation matrix in Table 5.3.

Additionally, associations between outcome variables and sociodemographic variables were explored with Spearman rank correlation analyses using a Bonferroni adjusted alpha level of 0.0025 (0.05/20) (Table 5.4). A longer duration of care recipients' cognitive impairment was associated with lower relationship

satisfaction for life partners ($p = 0.002$). Life partners' stress was higher when they provided more hours of care each week ($p < 0.001$).

For the sub-sample of 57 life partners and people with PRD, Spearman rank correlation analyses were performed using Bonferroni adjusted alpha level of 0.006 (0.05/8). The **duration of caregiving years** correlated with the partners' PD and cognitive impairment duration (both $p < 0.001$), H&Y stage ($p = 0.001$), SE-ADL ($p < 0.001$) and weekly care provision hours ($p = 0.002$) but not with UPDRDS-III ($p = 0.109$) (Table 5.5). There was also a significant negative association between **weekly care provision hours** and SE-ADL ($p = 0.001$) and a positive association between weekly hours of caregiving and PD duration ($p = 0.006$) but weekly care provision hours were not related to PD motor symptom severity (i.e. UPDRS-III and H&Y stage, $p > 0.006$).

Table 5.3 Spearman correlation analyses between outcomes (n = 136 life partners).

	RSS	ZBI	BRS	HADS-A	HADS-D	SF-12-PCS	SF-12-MCS	EQ5D-index	EQ5D-VAS	Rel.SS	DRS-inter.	DRS-strain	FCR-satisf.	FCR-resent.
ZBI	**-0.712*****													
BRS	**0.359*****	**-0.487*****												
HADS-A	**-0.432*****	**0.689*****	**-0.594*****											
HADS-D	**-0.553*****	**0.681*****	**-0.547*****	**0.760*****										
SF-12-PCS	0.030	-0.019	0.128	-0.054	-0.162									
SF-12-MCS	**0.494*****	**-0.635*****	**0.599*****	**-0.742*****	**-0.662*****	-0.157								
EQ-5D index	**0.281****	**-0.286****	**0.350*****	**-0.448*****	**-0.468*****	0.597***	**0.345*****							
EQ-5D VAS	**0.266****	**-0.279****	**0.345*****	**-0.374*****	**-0.391*****	**0.591*****	**0.299*****	**0.511*****						
Rel.SS	**-0.624*****	**0.872*****	**-0.505*****	**0.672*****	**0.694*****	**-0.014**	**-0.671*****	**-0.387*****	**-0.266****					
DRS-inter.	**0.351*****	-0.209	0.122	-0.126	-0.177	-0.093	0.145	0.096	0.001	-0.153				
DRS-strain	**-0.636*****	**0.710*****	**-0.330*****	**0.441*****	**0.487*****	0.055	**-0.455*****	-0.225	**-0.272****	**0.639*****	**-0.314*****			
FCR-satisf.	0.216	-0.210	0.077	-0.042	-0.103	-0.121	0.005	-0.128	-0.066	-0.112	**0.387*****	**-0.369*****		
FCR-resent.	**-0.612*****	**0.752*****	**-0.427*****	**0.605*****	**0.701*****	0.112	**-0.578*****	**-0.258****	-0.184	**0.748*****	-0.201	**0.569*****	-0.123	
FCR-anger	**-0.571*****	**0.598*****	**-0.320****	**0.464*****	**0.383*****	0.102	**-0.463*****	**-0.266****	-0.212	**0.584*****	**-0.428*****	**0.659*****	**-0.346*****	**0.546*****

Notes: ** $p < 0.003$, *** $p < 0.001$ (Bonferroni correction applied)

Abbreviations: BRS – Brief Resilience Scale; DRS – Dyadic Relationship Scale, positive interaction or negative strain sub-scale; EQ-5D – EuroQoL index score or visual analogue scale (VAS); FCR – Family Caregiving Role scale, caregiving satisfaction, resentment or anger sub-scale; HADS – Hospital Anxiety and Depression Scale, anxiety or depression sub-scale; Rel.SS – Relatives' Stress Scale; RSS – Relationship Satisfaction Scale; SF-12 – Short Form 12 Health Survey, physical health (PCS) or mental health (MCS) sub-scale; ZBI – Zarit Burden Interview.

Table 5.4 Spearman correlation analyses between outcomes and sociodemographic variables (n = 136 life partners).

	Age	Years of care provision	Weekly care provision hours	PD duration	Cognitive impairment duration
RSS	0.222	-0.163	-0.131	-0.178	**-0.264****
ZBI	-0.208	0.174	0.203	0.074	0.243
BRS	0.027	-0.094	-0.125	-0.004	-0.086
HADS-A	-0.121	0.102	0.079	-0.078	0.081
HADS-D	-0.108	0.127	0.220	0.039	0.109
SF-12-PCS	-0.234	-0.020	-0.106	-0.034	0.037
SF-12-MCS	0.224	-0.109	-0.162	0.052	-0.108
EQ5D-index	-0.076	-0.101	-0.196	-0.036	-0.085
EQ5D-VAS	-0.029	0.015	-0.180	0.044	0.026
Rel.SS	-0.209	0.214	**0.298*****	0.116	0.204
DRS-inter.	0.073	-0.007	0.136	-0.079	0.019
DRS-strain	-0.244	0.097	0.150	0.127	0.125
FCR-satisf.	0.068	-0.016	0.132	-0.132	-0.050
FCR-resent.	-0.102	0.218	0.248	0.098	0.190
FCR-anger	-0.196	0.153	0.096	0.106	0.099

Notes: ** $p < 0.0025$, *** $p < 0.001$ (Bonferroni correction applied)

Abbreviations: BRS – Brief Resilience Scale; DRS – Dyadic Relationship Scale, positive interaction or negative strain sub-scale; EQ-5D – EuroQoL index score or visual analogue scale (VAS); FCR – Family Caregiving Role scale, caregiving satisfaction, resentment or anger sub-scale; HADS – Hospital Anxiety and Depression Scale, anxiety or depression sub-scale; PD – Parkinson's disease; Rel.SS – Relatives' Stress Scale; RSS – Relationship Satisfaction Scale; SF-12 – Short Form 12 Health Survey, physical health (PCS) or mental health (MCS) sub-scale; ZBI – Zarit Burden Interview.

Table 5.5 Associations between caring duration and people with PRD outcomes (n = 57 life partners).

	Caring duration (y)	Weekly caring (h)	PD duration	Cognitive impairment duration	MoCA	H&Y	UPDRS-III	SE-ADL	NPI-total
Weekly caring (h)	**0.261****								
PD duration	**0.673*****	**0.236****							
Cogn. imp. duration	**0.411*****	0.123	**0.309*****						
MoCA	0.081	-0.264	-0.028	0.059					
H&Y	**0.449****	0.284	**0.415****	0.265	-0.203				
UPDRS-III	0.230	0.328	0.298	0.165	-0.298	**0.662*****			
SE-ADL	**-0.551*****	**-0.443****	-0.340	**-0.361****	0.330	**-0.636*****	**-0.657*****		
NPI total	0.194	0.314	0.232	0.297	-0.029	0.241	0.300	-0.359	
NPI-carer distress	0.084	0.175	0.185	0.164	-0.055	0.341	0.349	-0.236	**0.830*****

Notes: ** $p < 0.006$, *** $p < 0.001$ (Bonferroni correction applied)

Abbreviations: Cogn.imp. – cognitive impairment; H&Y – Hoehn & Yahr scale; MoCA – Montreal Cognitive Assessment; NPI – Neuropsychiatric Inventory; PD – Parkinson's disease; SE-ADL – Schwab & England Activities of Daily Living scale; UPDRS-III – Unified Parkinson's disease Rating Scale part III.

5.4.4.2 Comparisons of life partner outcomes according to the clinical syndrome

A one-way ANOVA revealed that care provision hours each week did not differ between the three clinical syndromes (*p > 0.05*) (Table 5.6). However, when PD-MCI was compared with the two dementia groups combined, the independent t-test showed that life partners of people with PDD and DLB devoted more hours to caregiving each week (m = 102.59, SD = 60.72) than life partners of people with PD-MCI (m = 76.74, SD = 64.03) [t(133) = -2.16, p = 0.033].

In terms of specific life partner outcomes, a one-way ANOVA showed that there were several statistically significant differences between PD-MCI, PDD and DLB (Table 5.6). Due to the unequal group sizes, a Hockberg's GT2 post hoc test was applied to determine which groups differed from one another, which revealed that life partners of people with PDD and DLB experienced more burden and resentment than life partners of people with PD-MCI (both p < 0.05). In addition, life partners of people with PDD experienced lower relationship satisfaction (p = 0.047), higher stress levels (p = 0.019), and less positive interaction with partner (p = 0.018) compared to life partners of people with PD-MCI, but these variables did not differ between PD-MCI and DLB groups. Conversely, life partners of people with DLB had higher levels of anxiety (p = 0.010) and lower levels of mental health (p = 0.024) than life partners of people with PD-MCI, but no difference was found between PD-MCI and PDD groups on these variables. There were no statistically significant differences between PDD and DLB groups on any of the studied outcomes (p > 0.05).

Taking into consideration that three variables (i.e. HADS-depression, FCR-satisfaction sub-scale, and care provision years) failed the assumption of homogeneity of variance (i.e. Levene's test p < 0.05), a Welch test was run, which revealed there were statistical differences for HADS-depression and care provision years among life partners of people with PD-MCI, PDD or DLB (Welch test p < 0.05). Therefore, I proceeded with ANOVA for these two variables but using a Games-

Howell post hoc test, which did not assume equal variances in the groups. As for the FCR-satisfaction sub-scale, a Kruskal-Wallis H test was run, which revealed no statistical differences between the disease groups ($p > 0.05$). A statistically significant difference in the HADS depression scores between the three disease groups was found as determined by one-way ANOVA ($F(2,133) = 9.94$, $p < 0.001$). A Games-Howell post hoc test revealed that life partners of people with PD-MCI had significantly lower depression scores than those caring for people with PDD ($m = 6.46$, $SD = 3.95$; $p < 0.001$) and DLB ($m = 6.96$, $SD = 4.45$; $p < 0.001$) but the scores did not differ in the PDD and DLB groups ($p > 0.05$). A one-way ANOVA also revealed that the amount of years that participants had provided care to the care recipients differed between the three clinical syndromes. A Games-Howell post hoc test determined that life partners of people with PDD provided care for longer ($m = 7.74$, $SD = 5.62$) than life partners of people with PD-MCI ($m = 4.68$, $SD = 3.35$; $p = 0.006$) and DLB ($m = 3.68$; $SD = 3.4$; $p = 0.000$).

Table 5.6 Outcomes for life partners of people with PD-MCI, PDD and DLB (n = 136).

	ANOVA					Post hoc test	
	m (SD)			F	p	p	
Diagnosis	**PD-MCI (n = 37)**	**PDD (n = 50)**	**DLB (n = 49)**	**df (2,133)**		**PD-MCI vs PDD**	**PD-MCI vs DLB**
Years caring	4.68 (3.35)	7.74 (5.62)	3.68 (3.43)	11.53	**0.000**	**0.006**‡	n.s.
Hours caring pw	76.74 (64.03)	106.77 (63.15)	98.32 (58.48)	2.55	0.082	n.s.	n.s.
RSS	28.68 (10.61)	22.48 (12.16)	24.22 (12.03)	3.07	**0.050**	**0.047**†	n.s.
ZBI	28.16 (14.19)	38.06 (14.00)	37.99 (16.78)	5.68	**0.004**	**0.009**†	**0.011**†
BRS	3.60 (0.76)	3.51 (0.78)	3.47 (0.86)	0.31	0.732	n.s.	n.s.
HADS-anxiety	5.73 (3.83)	7.88 (4.33)	8.65 (5.03)	4.69	**0.011**	n.s.	**0.010**†
HADS-depress.	3.46 (2.52)	6.46 (3.95)	6.96 (4.45)	9.94	**0.000**	**0.000**‡	**0.000**‡
SF-12-PCS	51.96 (10.23)	49.10 (10.62)	50.49 (11.23)	0.76	0.471	n.s.	n.s.
SF-12-MCS	48.28 (10.42)	44.87 (9.16)	42.28 (11.06)	3.63	**0.029**	n.s.	**0.024**†
EQ-5D index *	0.83 (0.19)	0.76 (0.24)	0.77 (0.19)	1.39	0.253	n.s.	n.s.
EQ-5D VAS *	78.03 (14.97)	75.22 (17.95)	73.54 (16.39)	0.77	0.465	n.s.	n.s.
Rel.SS	21.65 (9.59)	27.94 (10.85)	26.80 (10.77)	4.15	**0.018**	**0.019**†	n.s.
DRS-interaction	10.36 (3.57)	8.46 (3.03)	9.54 (2.87)	4.03	**0.020**	**0.018**†	n.s.
DRS-strain	4.38 (3.23)	6.04 (3.50)	5.64 (3.13)	2.85	0.061	n.s.	n.s.
FCR-resentment	2.38 (0.80)	2.92 (0.97)	2.91 (1.03)	4.25	**0.016**	**0.029**†	**0.035**†
FCR-anger	1.69 (0.73)	2.11 (0.86)	2.06 (0.83)	3.26	**0.042**	n.s.	n.s.

Notes: † - Hockberg's GT2 post hoc test; ‡ - Games-Howell post hoc test; *- winsorized.

Abbreviations: BRS – Brief Resilience Scale; DLB – Dementia with Lewy bodies; DRS – Dyadic Relationship Scale, positive interaction or negative strain sub-scale; EQ-5D – EuroQoL index or visual analogue scale (VAS); FCR – Family Caregiving Role scale; HADS – Hospital Anxiety and Depression Scale; IQR – interquartile range; n.s. – not significant; PDD – Parkinson's disease dementia; PD-MCI – Parkinson's disease and mild cognitive impairment; pw – per week; PwPRD – people with Parkinson's-related dementia; Rel.SS – Relatives' Stress Scale; RSS – Relationship Satisfaction Scale; SD – standard deviation; SF-12 – Short Form 12 Health Survey, physical health (PCS) or mental health (MCS) sub-scale; ZBI – Zarit Burden Interview.

5.4.5 Psychometric properties of the scales

Internal consistency was very good for all scales (Cronbach α > 0.8) except for DRS-positive interaction sub-scale (Cronbach α = 0.724), FCR caregiving satisfaction sub-scale (Cronbach α = 0.661) and FCR-anger sub-scale (Cronbach α = 0.729) (see Table 9). The completion rate was high for all scales (range 93.3-100%) and the total amount of missing data were very low (1%). The scales that had the most missing values were ZBI and DRS. Two sub-scales had either a floor or a ceiling effect: the EQ-5D index scale had a ceiling effect as 37 (27.2%) participants achieved the highest possible score in the scale (i.e. 1.000), and the FCR-anger had a floor effect as 33 (24.3%) respondents had the lower possible score in the scale (i.e. 1.0).

The high correlations between similar constructs (i.e. between RSS and DRS-positive interaction; among ZBI, Rel.SS, FCR-resentment and FCR-anger; among SF-12 mental health sub-scale, EQ-5D, HADS-depression and HADS-anxiety) indicated moderate to strong *convergent validity* between the scales. However, only ZBI and Rel.SS had a Spearman correlation coefficient above 0.8 suggesting that the convergent validity should be accepted for both scales. All sub-scales of the same scale were expected to have a strong correlation; however, only HADS anxiety and depression sub-scales had a Spearman correlation coefficient above 0.6. Other scales with multiple sub-scales had a moderate correlation (i.e. Spearman correlation coefficient between 0.4 and 0.59 for EQ-5D index and EQ-5D VAS; FCR-resentment and FCR-anger sub-scales), weak correlation (i.e. Spearman correlation coefficient between 0.20 and 0.39 for DRS-interaction and DRS-strain; FCR-anger and FCR-satisfaction sub-scales), or no correlation (i.e. Spearman correlation coefficient below 0.19 for SF-12 PCS and MCS; FCR-satisfaction and FCR-resentment sub-scales). These results suggest that the best psychometric properties were achieved for RSS, ZBI, BRS, Rel.SS, SF-12 and EQ-5D and these scales can be recommended to be used with life partners of people with PRD, whereas DRS and FCR achieved the lowest psychometric properties and are not favoured to be used with this population.

5.5 Discussion

5.5.1 General discussion

This study described the profile of life partners of people with PRD, compared life partner outcomes according to the care recipients' clinical syndrome (i.e. PD-MCI, PDD or DLB), and evaluated the psychometric properties of the scales in order to provide recommendations for future studies. This is the first study to explore the profile of life partners of people with PD-MCI, PDD and DLB collectively under the umbrella term of 'Lewy body spectrum disorders'. Exploring the demographic and clinical profile of life partners of people with PRD is important as a better understanding of these aspects can help with targeting support and tailoring interventions for this group.

A typical life partner who provided care to a person with PRD was a woman of 69.5 years of age having been married for over 45 years. People with PRD were mostly men aged about 73.5 years who had PD-MCI, PDD or DLB for a median of five years, which resembles to study populations in previous research (Cifu et al., 2006; Drutyte, Forjaz, Rodriguez-Blazquez, Martinez-Martin, & Breen, 2014; Hand et al., 2018; Lökk, 2008; Martinez-Martin et al., 2015; Peters et al., 2011). Notably, although the current study recruited only those participants who were spouses or partners of people with PRD, this is representative of a typical care partner of a person with PD or PRD in western countries as the majority of care partners are female spouses (Cifu et al., 2006; Hand et al., 2018; Lökk, 2008; Martinez-Martin et al., 2015; Peters et al., 2011).

The current study demonstrated that relationship dissatisfaction, burden, stress and feelings of resentment and anger were common among life partners, which resonate with earlier findings as stress, burden and quality of life among care partners of people with PD and PRD have been extensively studied (Grün, Pieri, Vaillant & Diederich, 2016; Lawson et al., 2018; Leroi et al., 2012a; Martinez-Martin et al., 2015; Mosley et al., 2017). However, relationship dissatisfaction, perceived

negative feelings (resentment) and resilience are new findings emerging from this study and appear to be under-researched in the field of PRD, despite numerous studies evaluating these constructs in other types of dementia (Croog, Burleson, Sudilovsky, & Baume, 2006; Evans & Lee, 2014; Parkinson, Carr, Rushmner, & Abley, 2017; Pozzebon et al., 2016). This is important as it could be hypothesised that care partner outcomes could be similar in PRD and other types of dementia but evidence suggests that care partners of people with PRD have higher rates of burden (Shin et al., 2012; Svendsboe et al., 2016), stress (Lee et al., 2013; Ricci et al., 2009), depression (Roland & Chappell, 2017), more tensions and arguments in the dyadic relationship (Davis, Gilliss, Deshefy-Longhi, Chestnutt, & Molloy, 2011) and lower abilities to live well (Wu et al., 2018) than care partners of people with AD and/or vascular dementia. This suggests care partners of people with PRD may have poorer health outcomes compared to care partners of people with non-PD type dementia, which has recently been confirmed in a large comparative study with people with various types of dementia and their care partners (Wu et al., 2018). Notwithstanding the high prevalence of negative outcomes, many life partners in the current study had good resilience, expressed satisfaction towards their caring role and had a relatively good quality of life, emphasising that both positive and negative outcomes within a care relationship can co-exist (Lawton et al., 1991). Furthermore, many life partners of people with PRD have accepted the situation they were in and learned to adapt and adjust to the challenging nature of the condition (Vatter et al., 2018a), despite the demands and stresses they faced. Frequently, care provision takes place within a long-term intimate relationship, and having a good relationship quality is important as it can protect against stressors and support care partners' quality of life (Goldsworthy & Knowles, 2008; Lawson et al., 2018); therefore, strengthening and supporting interpersonal relationships is central and should be the focus in the future studies.

The number of years that life partners in the current study had dedicated to care provision is comparable to earlier studies (Hand et al., 2018) but on average participants in the current study were providing fewer hours of care per day than care partners of people with PD in the Hand and colleagues' study (2018). This

difference could be explained in multiple ways: (1) the people with PD in Hand et al.'s study (2018) had a moderate to late stage PD and only included 10 people with PDD (8.7% of the total sample of 115 participants), (2) care partners in the current study may have underreported the care-related tasks as they may have been doing similar tasks premorbidly (i.e. cleaning, cooking, housework, etc), and (3) each care partner provided a subjective account of care provision hours which may vary greatly from day to day depending on their partners' symptoms and needs. Nevertheless, the care provision by over half of life partners in the current study exceeded 14 hours each day and over 100 hours each week which is significantly higher than the level found in care partners of people with dementia (i.e. between 3 and 11 hours per day; Brodaty & Donkin, 2009) and the rest of care partners in the UK (NHS Digital, 2017) emphasising the complexity of PRD as well as an immense commitment by care partners to take care of the people with PRD.

Several noteworthy differences in life partner outcomes according to the diagnostic category were found. As expected, life partners of people with PDD had provided care for more years than life partners of people with PD-MCI, and life partners of people with PDD and DLB were providing more hours of care each week than life partners of people with PD-MCI. A linear relationship was found between several variables and progression of cognitive impairment in PD. Once dementia in PD had emerged, life partners were more burdened, stressed, depressed, resentful, dissatisfied with the relationship and experienced fewer positive interactions with their partner compared to those whose partner had PD-MCI. Similarly to PDD, life partners of people with DLB had higher rates of depression, burden and feelings of resentment in comparison to life partners of people with PD-MCI. Importantly, life partners of people with DLB had higher anxiety levels and reported lower levels of mental health compared to life partners of people with PD-MCI, whereas these outcomes did not differ between PD-MCI and PDD groups, suggesting that specific clinical syndrome plays an important role in determining life partner outcomes.

The finding that life partners of people with DLB have high levels of anxiety and poor mental health appears to be novel despite comparative studies demonstrating

that these care partners have significantly more burden (Svendsboe et al., 2016) and stress (Lee et al., 2013; Ricci et al., 2009) compared to care partners of people with other types of dementia. The impact of DLB on life partners can be more profound as the speed of onset is faster, intensity of symptoms and levels of fluctuation higher, and impairments in certain areas of cognitive functioning greater than in PDD (Camicioli & Fisher, 2005; Park et al., 2011). Interestingly, however, this study found no differences in life partner outcomes between people with PDD and DLB. This could imply that both diseases have a similar impact on life partners, which could be due to the two diseases having a clinically similar symptom presentation in terms of cognitive, psychiatric and motor symptoms and share underlying pathology (Aldridge et al., 2018; Friedman, 2018; Jellinger & Korczyn, 2018; McKeith et al., 2004; McShane, 2008; Noe et al., 2004; Thomas et al., 2005).

Finally, the psychometric properties of the scales were evaluated. The scales that have been commonly used among care partners of people with PD and PRD (i.e. ZBI, Rel.SS, SF-12 and EQ-5D) appeared to be psychometrically robust instruments with good internal reliability and validity. In addition, other scales which had not been used among care partners of people with PRD before (i.e. RSS, BRS) had good psychometric properties and could be used in future studies to evaluate outcomes in care partners. However, lesser known scales (such as DRS and FCR), which to my knowledge were evaluated for the first time with life partners of people with PRD, had low psychometric properties, low convergent validity, more missing values (i.e. DRS) and a floor effect (i.e. FCR) and the researchers using these scales in future studies may need to be cautious. The EQ-5D measure, which has been used previously to evaluate quality of life of people with PRD (Shin et al., 2012) appeared to be suitable to use in this population but the presence of the ceiling effect may make the interpretation of results more difficult. Evaluating psychometric properties of the instruments is important to determine that the chosen scales are valid, accurate and reliable and the emerging findings can be trusted.

5.5.2 Methodological strengths

The current study recruited spouses and life partners of people with Lewy body spectrum disorders, namely PD-MCI, PDD and DLB, and is the largest study to date of life partners of people with PRD. This study is the first to describe the demographic and clinical profile in a sample of life partners of people with PRD that also compares outcomes according to the diagnostic category in the context of PRD. Earlier studies have often compared care partner outcomes of people with PDD and/or DLB to life partners of people with other types of dementia such as AD, vascular dementia or frontotemporal degeneration but have not studied the PD-MCI group. Including care partners of people with PD-MCI is important because once cognitive impairment has emerged in PD the likelihood of progressing to dementia increases significantly and researchers have illustrated that already at the PD-MCI stage, care partners feel stressed and burdened. Therefore, one of the key strengths in the study was the analyses and comparison of the results in the three PD-related cognitive impairment types.

This study also included a variety of scales which allowed a detailed description of life partner outcomes and a better understanding how a specific clinical syndrome within the context of PD and cognitive impairment affected the life partner. Additionally, the evaluation of the psychometric properties of the scales, which is not commonly reported in the majority of studies, strengthened credibility and accuracy of the findings, and also highlighted the instruments that may not be appropriate to use in future studies with life partners of people with PRD. A detailed description of the scales also increases transparency and clarity about study findings and supports the selection of the chosen statistical analyses.

5.5.3 Limitations

The present study is not without its limitations. First, as this study combined participants recruited through two different routes, the disease-specific aspects, such as disease severity, stage, functional ability, severity and frequency of

neuropsychiatric symptoms, and cognitive impairment test scores, could not be elicited for those people with PRD whose life partner participated via the postal questionnaire study, which precluded the exploration of the impact of disease-specific aspects on life partners.

Second, including participants from the INVEST study introduced a sample selection bias as some participants were excluded due do not being a partner or a spouse of a person with PRD and some participants were contacted twice (first for the purposes of the INVEST study and then for the postal questionnaire study if they had screen-failed or refused participation in the INVEST study), which indicated a non-random sample as not all participants were equally likely to be included in the study. Furthermore, self-selection bias was present in participants recruited through the postal questionnaire study as participants decided whether they would like to take part in the study or not. The biases could have been overcome by recruiting all participants through one route (i.e. either postal questionnaire or INVEST study only).

Third, this study did not include an age-matched cohort as a control group, which limits the possibility of comparing the outcomes of life partners of people with PRD with the outcomes of the general population. Earlier comparative studies found that care partners of people with PD have lower levels of mental health (Aarsland et al., 1999a; Peters et al., 2011) and health-related quality of life (Martinez-Martin et al., 2008) in comparison to the general population but it remains unknown how other outcomes, such as relationship satisfaction, stress, burden, resilience, anxiety, depression and physical health, differ between life partners and general population. Therefore, it would be important to include a comparative group in addition to life partners of people with PRD in a future study.

Fourth, although this is the most extensive study with life partners of people with PRD to date, a sample size of at least 200 participants is recommended (Frost et al., 2007) to accurately report on the reliability and validity of the instruments. However, this study provided preliminary evidence of the psychometric properties

of the scales which could be used in care partners of people with various types of dementia.

Fifth, this study explored a limited number of psychometric properties (i.e. internal consistency, convergent validity, response rate, floor and ceiling effects) and did not evaluate other types of reliability (i.e. inter-rater reliability, test-retest reliability) and validity (i.e. content, construct and criterion validities) (Cook & Beckman, 2006; Cordier et al., 2017). Although a more thorough investigation of additional psychometric properties would have further increased knowledge about the accuracy and appropriateness of the scales in this population, it was not the main aim of the study; rather, the current study provided preliminary information about the use of these measures, most of which were novel in this population. Next, life partners' health history was not explored in detail and this could have expanded knowledge regarding their own physical and mental health needs which impact on their ability to provide care to their partners.

Finally, since this was a cross-sectional study using self-reported measures, it should be acknowledged that the information provided by life partners is subjective and could have been biased, overestimated or underestimated depending on how life partners felt at the time of the assessment. Applying a longitudinal design could support the accuracy of the findings as well as provide information regarding inter-rater reliability.

5.5.4 Future directions

A number of recommendations could be made for a future study. The subsequent study could include people with PD-MCI, PDD and DLB, dyads with various care relationship types (i.e. spouses, partners, adult children, other family members) and people with different cultural backgrounds. The study could also recruit a larger sample of care partners of people with PRD to increase external validity and evaluate psychometric properties of various measures in-depth. A thorough literature review summarising the role and profile of care partners of people with

PRD could provide an accurate depiction of this population. In addition, including a wide assessment battery and conducting a clinical examination of both people with PRD as well as their care partners would allow a better understanding of the health and well-being of each member of the dyad.

5.6 Conclusion

This is the first and largest study to date describing the profile of life partners of people with Parkinson's-related dementia. Importantly, this study highlighted that life partners of people with PDD and DLB have high rates of burden, stress, relationship dissatisfaction and negative feelings as well as poor levels of mental health. Focusing on supporting dyadic relationships should be the aim of future research as good relationships can support the well-being and quality of life of both partners and potentially delay institutionalisation of the person with PRD, which has cost saving implications.

CHAPTER 6: Burden in life partners of people with Parkinson's-related dementia (Study 2)

The study described in this chapter has been published:

Vatter, S., McDonald, K. R., Stanmore, E., Clare, L., & Leroi, I. (2018b). Multidimensional care burden in Parkinson-related dementia. *Journal of Geriatric Psychiatry and Neurology, 31(6)*, 319-328.

6.1 Introduction

Care provision in Parkinson's disease (PD) is considered challenging as it involves providing help with multiple aspects of the disease, including motor, cognitive and psychiatric symptoms and oversight of complex treatment regimens. The demands of the caring role can be both physically challenging and mentally exhausting for life partners (Roland et al., 2010; Tan et al., 2012), resulting in stress, strain and burden (Carter et al., 2008; Leiknes et al., 2015; Lökk, 2008; Martinez-Martin et al., 2008, 2015; Mosley et al., 2017; Whetten-Goldstein et al., 1997). Consequently, life partners can have increased anxiety and depression (Martinez-Martin et al., 2008; Schrag et al., 2006), reduced life satisfaction (Aarsland et al., 1999a), lower quality of life (Lawson et al., 2017; Leroi et al., 2012; Martinez-Martin et al., 2008) and higher rates of mortality (Schulz & Beach, 1999). In the literature life partners have been referred to as 'the hidden patients' (Fengler & Goodrich, 1979) highlighting the importance of focusing on this population.

One of the most extensively researched constructs related to care provision is 'caregiver burden', which is defined as "the extent to which caregivers perceive their emotional or physical health, social life, and financial status as suffering as a result of caring for their relative" (Zarit et al., 1986, p. 261). Studies have concluded that burden increases with the emergence of cognitive impairment and dementia in PD (Cifu et al., 2006; Leroi et al., 2012a; Martinez-Martin et al., 2015), and is higher amongst care partners of people with Parkinson's disease dementia (PDD) than those of people with AD due to advanced functional impairment in people with PDD

(Shin et al., 2012). Use of the term 'burden' is becoming less popular, as the provision of care for a loved one may not be experienced as a burden and strain but as a marital contract, commitment and moral responsibility (Kilgariff & Grant, 2016). The term, however, remains common in literature and relevant to the topic at hand, therefore, it will be used in this chapter for consistency with earlier research.

The most extensive measure of burden amongst life partners of people with PD (Leiknes et al., 2015) is the Zarit Burden Interview (ZBI; Zarit et al., 1980). The ZBI, a 22-item scale, assesses life partners' physical, emotional and socio-economic status in regards to care provision (Zarit et al., 1980) and is deemed a reliable and valid scale with good psychometric properties (Hagell, Alvariza, Westergren & Arestedt, 2017). Despite the fact that the ZBI is typically examined as a unitary construct, studies have suggested that ZBI is multifactorial with 'personal strain' and 'role strain' as common factors (Bedard et al., 2001; Branger, O'Connell & Morgan, 2016; Harding et al., 2015; Zarit & Zarit, 1990). Many researchers have confirmed the multidimensionality of the ZBI in conditions other than PD or Parkinson's-related dementia (PRD) with solutions of two-factor (Branger et al., 2016), three-factor (Ankri, Andrieu, Beaufils, Grand & Henrard, 2005; Leggett, Zarit, Taylor & Galvin, 2011; Oh & Kim, 2018; Pillemer, Davis & Tremont, 2018; Siegert et al., 2010; Smith, George & Ferriera, 2018; Springate & Tremont, 2014), four-factor (Cheah, Han, Chong, Anthony & Lim, 2012; Cheng, Kwok & Lam, 2014), five-factor (Lu, Wang, Yang, & Feng, 2009; Tang et al., 2017) and six-factor (Torres, Hoff, Padovani & Ramos-Cerqueira, 2012) models. Frequently the three-factor solutions have been named as 'impact on caregiver's[3] personal and social life', 'feelings of frustration, embarrassment and/or anger', and 'guilt or self-criticism' (Ankri et al., 2005; Oh & Kim, 2018; Smith et al., 2018; Springate & Tremont, 2014). The variance in the factor solutions suggests that culture (Sousa et al., 2016) and clinical syndrome (Harding et al., 2015) play an important role in care partners' burden.

[3] The term 'carer' are 'caregiver are common in the UK and USA, respectively; however, the use of the term 'care partner' has become more acceptable, which will be used throughout the chapter.

Many authors have confirmed that burden is a multidimensional construct which is supported with the definition of burden provided by Zarit *et al.* (1986). One of the best techniques to examine the dimensionality of a scale is by undertaking an exploratory factor analysis (EFA) which groups meaningful variables together into distinct factors (Worthington & Whittaker, 2006). An EFA combines quantitative methodology with an inductive approach to explore the number of factors that underlie a set of variables, describe the features of the emerging factors and evaluate the suitability of factors through examination and revision in a dynamic manner (Worthington & Whittaker, 2006). In short, the purpose of an EFA is to simplify the data, capture meaningful patterns in the data and reduce the data into smaller sets of constructs. As burden is a latent variable (i.e. not observed directly and measured as a set of multiple items), researchers have applied a factor analysis technique to explore the latent dimensions of the burden (measured by ZBI) in different disease areas. Specifically, factor analyses of ZBI have been conducted with care partners of people with memory impairment (Cheah et al., 2012), dementia (Ankri et al., 2005; Smith et al., 2018; Springate & Tremont, 2014), and dementia with Lewy bodies (DLB; Leggett et al., 2011). EFA studies of the ZBI have been undertaken across various countries but only one study has been conducted in the UK where authors found that the UK care partners had higher burden levels in comparison to other countries (Smith et al., 2018). Despite the fact that there are several factor analytic studies of the ZBI, the factor structure of the ZBI in life partners of people with PRD remains unknown. Evidence has shown that transitioning from PD to mild cognitive impairment (PD-MCI) and dementia (PDD) is accompanied by a significant decline in function and marked increase in negative impact on care but the aspects that exacerbate care provision are less understood.

Given the multidimensionality of burden as measured by the ZBI, the low number of studies with a UK sample and the lack of understanding as to what constitutes burden in life partners of people with PRD, there is an opportunity to conduct an exploratory factor analysis of ZBI in spouses and life partners of people with PD-MCI, PDD and DLB.

6.2 Aims

The aims of the current study were to (1) explore the factor structure of the Zarit Burden Interview (Zarit et al., 1980) in spouses and life partners of people with PRD, and (2) examine the associations and predictors between the emerging factors and the demographic and clinical features.

An exploratory factor analysis method was deemed appropriate for the purposes of this study as EFA explores the relationships between the variables and the number of factors in a new sample (Worthington & Whittaker, 2006). It was hypothesised that a three-factor-solution would emerge according to similar research conducted with care partners of people with dementia (Ankri et al., 2005; Leggett et al., 2010; Smith et al., 2018; Springate & Tremont, 2014).

6.3 Methods

6.3.1 Research design

This was a cross-sectional study design. Full ethical approval for the study was granted through a Substantial Amendment to the INVEST project by the Yorkshire & the Humber – Bradford Leeds Research Ethics Committee (reference number: 15/YH/0531) on 09/06/2017 (see Chapter 4, Section 4.2.8).

6.3.2 Participant sample

Spouses and life partners of people with PRD were recruited through two different routes: (1) a face-to-face home-based interview conducted by a researcher (undertaken as part of the baseline assessment visit in the INVEST study), and (2) a UK-wide postal questionnaire study. Participants were eligible to participate if they were a partner or a spouse of a person diagnosed with PD-MCI, PDD or DLB, and if they lived together. Participation of people with PRD was not required in this study;

however, participants who took part in the postal questionnaire study provided information about their partners with PRD.

The recruitment target was a minimum of 100 participants as per Gorsuch's (1983) and Kline's (1994) recommendations on the minimum required sample size for a factor analysis study. Several authors have also suggested minimum participant-to-item-ratios with Cattell (1978) proposing three to six participants per variable, Gorsuch (1983) recommending at least five participants per variable and Comrey and Lee (1992) suggesting at least five to ten participants per variable. The proposed ratio of sample size *(n)* to the number of variables *(p) (n:p)* by Gorsuch (1983) and Cattell (1978) is in line with the current study exploring a factor structure of a 22-item ZBI scale (Zarit et al., 1980), with five participants per item bringing the sample size to 110. Furthermore, it was anticipated that missing data would occur in 10% of the cases; therefore, a sample size of at least 121 participants was planned.

In order to test individual predictors in multiple regression analyses, a sample size of $\geq 104 + m$ was recommended, where m was the number of independent variables (Green, 1991; VanVoorhis & Morgan, 2007), which satisfied the rule of recruiting a minimum of 121 participants. The number of predictors was determined following the exploratory factor analysis.

In home-based assessments, the baseline data of 57 life partners, who participated in the INVEST study, could be elicited. Thus, to reach the sample size of 121 life partners, 64 additional participants were required to complete the postal survey. Taking into account the previous response rates of 39-77% of recruitment via postal questionnaire studies (Jakobsen, Poulsen, Reiche, Nissen, & Gundgaard, 2011; Laakkonen et al., 2008; McRae et al., 2009; Morley et al., 2013), it was estimated that the response rate in the current study would be about 50%.

6.3.3 Recruitment

Participant recruitment and the recruitment procedure for the INVEST study, from which the cross-sectional baseline data of life partners originates, is described in Chapter 4, Sections 4.2.2. Additionally, the recruitment of life partners is described in Chapter 5, Section 5.3.2.

6.3.4 Procedure

Life partners completed the quantitative measures during the home-based assessments or via the postal questionnaire (described in Chapter 5, Sections 5.3.2).

6.3.5 Measures

All participants were asked to fill in an assessment pack consisting of rating scales covering physical and psychological health, relationship satisfaction, resilience and feelings related to care provision. Specifically, participants completed the following assessments:

- Zarit Burden Interview (ZBI; Zarit et al., 1980),
- Relationship Satisfaction Scale (RSS; Burns, 1983),
- Relatives' Stress Scale (Rel.SS; Greene et al., 1982),
- Dyadic Relationship Scale (DRS; Sebern & Whitlatch, 2007),
- Family Caregiving Role scale (FCR; Schofield et al., 1997),
- Short Form 12 Health Survey (SF-12; Ware et al., 1996),
- Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983),
- EuroQoL-5D-3L (EQ-5D; The EuroQol group, 1990) and
- Brief Resilience Scale (BRS; Smith et al., 2008).

Furthermore, severity of care recipients' Parkinson's disease (measured by Hoehn & Yahr stage, H&Y, Hoehn & Yahr, 1967) and neuropsychiatric symptoms (measured by the Neuropsychiatric Inventory, NPI, Cummings et al., 1994) were obtained in home-based assessments. All measures are described in detail in Chapter 4, Section

4.2.4.1, and the psychometric properties of these scales, are described in Chapter 5, Study 1.

In addition to the scales, demographic information such as age, gender, education, ethnicity, marital status, relationship duration and living status of both partners as well as partners' diagnosis, the onset year of PD or DLB symptoms and cognitive impairment, and care provision duration in years and weekly hours by life partners was elicited through the questionnaires.

For the exploratory factor analysis only the ZBI scale was used. The **Zarit Burden Interview** (ZBI, Zarit et al., 1980) is a 22-item self-report scale assessing burden in the caregiving life partner (for example: "Do you feel that your partner is dependent upon you?"). The items on the ZBI were scored on a 5-point Likert scale from 0 (never) to 4 (nearly always), whereby a higher score indicated a higher level of burden experienced by the life partner. Possible scores were between 0 (minimum) to 88 (maximum) and the cut-off scores of ZBI were 0 – 20 for little or no burden, 21 – 40 indicating mild to moderate burden, 41 – 60 showing moderate to severe burden and 61 – 88 illustrating severe burden (Zarit, Orr, & Zarit, 1985). Earlier studies conducted with caregiving partners of people with dementia have found the internal reliability (Cronbach α) to be above 0.90 (Smith et al., 2018; Springate & Tremont, 2014). The ZBI item (number 22: "Overall, how burdened do you feel in caring for your partner?"), which evaluates burden globally, was removed from the EFA due to its high correlation with the other items on the scale (Lu et al., 2009).

6.3.6 Analyses

All statistical analyses were conducted with IBM Statistical Package for Social Sciences software for Windows, version 23.0 (SPSS, IBM Corp, 2015). Any missing data were deleted listwise. The statistical analyses included descriptive (mean, range, standard deviations, %, internal reliability) and inferential statistics (mean comparison with ANOVA, exploratory factor analysis of the ZBI, multiple regression

analyses). Data were examined for normality of distribution with the Shapiro-Wilk test and with visual inspection of the histograms. The significance level for results was set at $p<0.05$.

Prior to conducting EFA, descriptive statistics and assumption tests were carried out to verify whether the dataset was suitable for conducting an EFA. The first step was to observe the mean values and standard deviations of each item on the ZBI to verify the absence of outliers. Next, a correlation matrix was undertaken to examine the relationship pattern amongst the variables (Yong & Pearce, 2013). If any of the variables had high correlation coefficients (i.e. $r < \pm 0.90$), they were removed as they showed multicollinearity (Yong & Pearce, 2013). Next, Bartlett's test of sphericity and the Kaiser-Meyer-Olkin measure were carried out to confirm the suitability of the data (Howard, 2016; Yong & Pearce, 2013). Bartlett's test of sphericity evaluated whether the correlation matrix in the current study was distinct from the identity matrix. If a difference was found ($p < 0.05$), it indicated that the collected data were suitable and valid for structure detection and there were patterned relationships. The Kaiser-Meyer-Olkin measure of sampling adequacy (recommended to be above 0.50) measured whether the sample was adequate for EFA (Howard, 2016; Kaiser, 1974). The Kaiser-Meyer-Olkin statistic varies between 0 and 1 and the higher the value the more it indicates that a factor analysis should reveal reliable factors (Field, 2013). If the aforementioned assumption tests were met, an EFA was feasible to conduct.

The EFA was conducted using principal axis factoring, which is a common and often preferred method that produces accurate solutions (de Winter & Dodou, 2012; Howard, 2016; Osborne, 2014). Next, rotation was applied. Rotation creates a pattern of loadings where high loadings are maximised and low loadings are minimized resulting in an interpretable, clear and understandable output (Costello & Osborne, 2005; Osborne, 2014; Yong & Pearce, 2013). In order to determine whether to conduct an oblique (factors are correlated) or orthogonal (factors are uncorrelated) rotation for the EFA, a preliminary factor analysis was run with oblique rotation to observe the correlations between the factors. If the majority of

correlations between the factors were above 0.32, an oblique rotation would be used in the EFA (e.g. direct oblimin or promax). However, if the factors were uncorrelated an orthogonal method (e.g. varimax or quartimax) for rotation would be used. Previously various researchers, who have undertaken an EFA of the ZBI have applied either oblique or orthogonal rotations (Ankri et al., 2005; Cheah et al., 2012; Leggett et al., 2010; Oh & Kim, 2018; Smith et al., 2018); thus, a decision in this study was made according to the correlations between the factors.

The number of factors was determined by eigenvalue scores that were greater than one (the Kaiser criterion; Kaiser, 1960) and inspection of the scree plot, which would explain a sufficient degree of variance. All items were reviewed to verify whether they had loaded onto any of the factors, whether they had a desirable loading equal or above 0.4 (recommended by Hinkin, 1995; Howard, 2016) and whether items had loaded on several factors. Following the extraction, rotation and verification of each item, the factors were named using previous literature and qualitative data analysis as a guide. Then, the reliability of each factor was calculated with Cronbach's alpha to determine whether each factor was reliable, i.e. Cronbach's $\alpha > 0.7$. If any of the factors appeared to have low reliability scores, items in the factor were reviewed to verify the item(s) that caused the low reliability score, these were removed, and the reliability test was re-run.

Once the factors were determined and named, multiple linear regression analyses were run to observe the predictors of each factor. Before running multiple linear regression analyses, the assumption tests were carried out through statistical tests and visual inspection of graphics which are described in Appendix H.

6.4 Results

6.4.1 Participant recruitment and response rate

A total of 136 life partners of people with PRD participated in the study. In route one, 57 life partners, who met the eligibility criteria for the current study, were

included from the INVEST study. In route two, a total of 186 postal questionnaires were posted out and 80 life partners returned the questionnaire (response rate 43%). From 80 completers, one participant was excluded due to their spouse living in a care home.

6.4.2 Characteristics of life partners and people with PRD

All couples lived together and the majority (94.9%) of life partners were married (mean relationship duration 42.27, SD = 13.60 years) and women (85.3%). Average age of life partners was 69.44 (SD = 7.62) years and 89.7% of them were white British. With regard to education, 61.8% of life partners were educated above compulsory education (e.g. had a further education qualification or a university degree). On average, participants had provided care for 5.51 (SD = 4.73) years and were currently providing care for 95.66 (SD = 62.91) hours a week.

The people with a diagnosis of PRD were mostly male (85.3%), white British (90.4%), with a mean age of 73.51 (SD = 6.48). Of them, 55.9% had completed further education or higher education. Thirty-seven people had a diagnosis of PD-MCI, 50 of PDD and 49 of DLB. The average disease duration of Parkinson's disease (including DLB diagnosis in the absence of PD diagnosis) and cognitive impairment was 7.14 years (SD = 6.52) and 4.59 (SD = 3.86) years, respectively. The characteristics of participants and their partners with PRD are provided in Table 6.1.

Table 6.1 Participant characteristics (n = 136 life partners).

Characteristics	Life partners	People with PRD
Gender, female n (%)	116 (85.3)	20 (14.7)
Age, mean years (SD)	69.44 (7.62)	73.51 (6.48)
Ethnicity, white British n (%)	122 (89.7)	123 (90.4)
Relationship status, n (%)		
Married	129 (94.9)	
Cohabiting	7 (5.1)	
Relationship duration, mean years (SD)	42.27 (13.60)	
Living status, n (%)		
With spouse/partner	134 (98.5)	
With spouse/partner + other family	2 (1.5)	
Education, n (%)		
School leaver at the age of 14-16 years	41 (30.1)	54 (39.7)
School leaver at the age of 17-18 years	11 (8.1)	6 (4.4)
Further education	34 (25.0)	36 (26.5)
Higher education (university degree)	50 (36.8)	40 (29.4)
Age left education, mean years (SD)	18.28 (4.78)	18.32 (5.19)
Diagnosis, n (%)		
PD-MCI		37 (27.2)
PDD		50 (36.8)
DLB		49 (36.0)
Duration of PD or DLB, mean years (SD)		7.14 (6.52)
Duration of MCI or dementia, mean years (SD)		4.59 (3.86)
Care duration, mean years (SD)	5.51 (4.73)	
Care hours per week, mean (SD)	95.66 (62.91)	

Abbreviations: DLB – Dementia with Lewy bodies; PD – Parkinson's disease; PDD – Parkinson's disease dementia; PD-MCI – Parkinson's disease and mild cognitive impairment; PRD – Parkinson's-related dementia; SD – standard deviation

6.4.3 Descriptive analyses of the Zarit Burden Interview (ZBI)

The mean score on the ZBI scale was 35.51 (SD = 15.40), median 35.00 and range 2 to 74. The items were normally distributed according to the Shapiro-Wilk test (p = 0.637) and the internal consistency reliability of the ZBI scale was excellent (Cronbach's α = 0.92). Missing items on the ZBI occurred in nine participants and their data were excluded listwise. The mean scores and SD's of each item revealed no abnormalities or outliers. Twenty (15.7%) life partners experienced little burden or no burden, 58 (45.7%) had mild to moderate burden, 39 (30.7%) had moderate to severe burden and 10 (7.9%) had severe burden. A one-way ANOVA comparing

the effect of clinical syndrome (PD-MCI, PDD, DLB) on the scores for the ZBI in life partners showed that there was a significant effect of clinical syndrome on the ZBI scores at the $p<0.01$ level [$F(2,124)=5.79$, $p = 0.004$] showing that burden was significantly lower in PD-MCI (m = 29.15, SD = 15.19) than in PDD (m = 40.84, SD = 14.23) and DLB (m = 38.50, SD = 17.35).

Higher burden in life partners was associated with lower relationship satisfaction (RSS), higher stress (Rel.SS), lower mental health score (SF-12), higher anxiety and depression (HADS), stronger feelings of resentment (FCR), higher negative strain (DRS), lower resilience (BRS) (all at $p<0.001$) and lower health-related quality of life (EQ-5D), lower positive interaction with the partner (DRS) and longer duration of partners' cognitive impairment (all at $p<0.01$) (Table 6.4). In addition, among the sub-sample of participants who were recruited through face-to-face home-based interview, data were available for people with PRD which showed that higher disease stage (H&Y) ($p<0.001$) and higher neuropsychiatric symptom score ($p<0.01$) was associated with higher burden in life partners.

6.4.4 Exploratory factor analysis of the ZBI

The data satisfied the Kaiser-Meyer-Olkin measure of sampling adequacy (= 0.882) and Bartlett's test of sphericity ($\chi^2 = 1340.248$, df = 210, $p < 0.0001$). A preliminary factor correlation matrix showed that the factors were not strongly correlated; therefore, an orthogonal method for rotation was applied. Principal axis factoring with varimax rotation was conducted which revealed a five-factor-solution with eigenvalues greater than 1 that accounted for 65.61% of the total variance (see Table 6.2). All factors were retained as they explained the highest percentage of the total variance. The internal reliability of each factor was good (Cronbach's α between 0.773 – 0.845). Factor five consisted of only two items; however, this corresponded with the findings from earlier research (Cheah et al., 2012; Cheng et al., 2014; Oh & Kim, 2018; Siegert et al., 2010; Tang et al., 2017) and the factor was preserved. Item 7 *("Are you afraid about what the future holds for your relative?")*

did not load onto any factors as it had a factor loading below 0.40 (Hinkin 1995; Howard, 2016) and was excluded from the model.

The five factors were as follows:

(1) *'Social and psychological constraints'* (items 1, 4-6, 9, 13, accounting for 39.33% of the variance) described the change in life partners' relationship with their friends as a result of care provision and the associated feelings (e.g. embarrassment, anger, strain),

(2) *'Personal strain'* (items 3, 10, 11, 15, accounting for 8.18% of the variance) represented the impact of providing care on the life partner (e.g. health problems, lack of privacy),

(3) *'Interference with personal life'* (items 2, 8, 12, 14, 17, accounting for 6.93% of the variance) illustrated the limitations to the life partners' life that had resulted from care provision (e.g. lack of time for self, limited social participation),

(4) *'Concerns about future'* (items 16, 18, 19, accounting for 5.80% of the variance) depicted fear and uncertainty in regards to care provision (e.g. inability to provide care, getting rid of the caring responsibility),

(5) *'Guilt'* (items 20, 21, accounting for 5.37% of the variance) reported the life partners' self-critical perception of their role (e.g. doing more, doing a better job in caring).

Table 6.2 Item descriptive statistics and standardised factor loadings of the ZBI (principal axis factoring, varimax rotation)*, loadings ≥ 0.400, n = 127 life partners.

		Factors (number of items)				
ZBI items	**Mean (SD)**	**1** (6)	**2** (4)	**3** (5)	**4** (3)	**5** (2)
Eigenvalue	N/A	8.26	1.72	1.45	1.22	1.13
% of variance	N/A	39.33	8.18	6.93	5.80	5.37
Internal consistency (Cronbach's α)	N/A	0.81	0.77	0.85	0.82	0.79
13. Do you feel uncomfortable having your friends over because of your relative?	0.87 (1.16)	0.664				
4. Do you feel embarrassed about your relative's behaviour?	0.98 (0.96)	0.607				
6. Do you feel that your relative currently affects your relationship with other family members?	1.01 (1.10)	**0.578**	0.408			
9. Do you feel strained when you are around your relative?	1.58 (0.10)	**0.542**	0.428			
1. Do you feel that your relative asks for more help than he or she needs?	1.35 (1.04)	0.477				
5. Do you feel angry when you are around your relative?	1.22 (0.92)	0.471				
15. Do you feel that you don't have enough money to care for your relative, in addition to the rest of your expenses?	1.14 (1.30)		0.596			
3. Do you feel stressed between caring for your relative and trying to meet other responsibilities for your family or work?	1.95 (1.17)		0.591			
10. Do you feel that your health has suffered because you are caring for your relative?	1.31 (1.17)	0.461	**0.578**			
11. Do you feel that you don't have as much privacy as you would like, because of your relative?	1.32 (1.27)		0.477			
2. Do you feel that, because of the time you spend with your relative, you don't have enough time for yourself?	2.30 (1.21)			0.691		
8. Do you feel that your relative is dependent upon you?	3.56 (0.80)			0.657		
12. Do you feel that your social life has suffered because you are caring for your relative?	2.28 (1.23)			0.578		
17. Do you feel that you have lost control of your life since your relative's illness?	1.81 (1.29)		0.446	**0.512**		
14. Do you feel that your relative seems to expect you to take care of him or her, as if you were the only one he or she could depend on?	2.34 (1.37)			0.442		
16. Do you feel that you will be unable to take care of your relative much longer?	0.94 (1.02)				0.845	
19. Do you feel uncertain about what to do about your relative?	1.26 (1.06)				0.614	
18. Do you wish that you could just leave the care of your relative to someone else?	0.94 (1.11)				0.580	
20. Do you feel that you should be doing more for your relative?	1.28 (1.16)					0.900
21. Do you feel that you could do a better job in caring for your relative?	1.37 (1.13)					0.715
7. Are you afraid about what the future holds for your relative?	2.77 (1.05)	–	–	–	–	–
22. Overall, how burdened do you feel in caring for your relative?†	1.91 (1.31)					

Notes: *Bold type indicates factor to which items are allocated; † item not included in factor analysis; – item removed due to poor loading; Factor 1 – 'Social and psychological constraints'; Factor 2 – 'Personal strain'; Factor 3 – 'Interference with personal life'; Factor 4 – 'Concerns about future', Factor 5 – 'Guilt'.

6.4.5 Correlation analyses of the ZBI and factors

All five factors correlated with the total ZBI score at the $p < 0.001$ level but factor 5 had the lowest correlation coefficient ($r = 0.41$). The strongest correlations between factors were found between factors 1 and 2 ($r = 0.67$), factors 2 and 3 ($r = 0.67$), factors 1 and 3 ($r = 0.64$) and factors 3 and 4 ($r = 0.62$) (Table 6.3). Item 22 correlated with the total ZBI score and all five factors.

Table 6.3 Pearson correlations between the ZBI and the factors (n = 127 life partners).

		Factors					
	ZBI	**1**	**2**	**3**	**4**	**5**	**Item 22**
Factor 1	0.85***	1.00					
Factor 2	0.85***	0.67***	1.00				
Factor 3	0.86***	0.64***	0.67***	1.00			
Factor 4	0.75***	0.55***	0.54***	0.62***	1.00		
Factor 5	0.41***	0.27**	0.24**	0.23**	0.30***	1.00	
Item 22	0.74***	0.59***	0.53***	0.64***	0.67***	0.26**	1.00

Notes: ** $p < 0.01$, *** $p < 0.001$

The scores of all five factors negatively correlated with relationship satisfaction (RSS), mental health score (SF-12) and resilience (BRS), and positively correlated with stress (Rel.SS), anxiety and depression (HADS), resentment sub-scale on the Family Caregiving Role (FCR) scale, negative strain sub-scale on the Dyadic Relationship Scale (DRS) and care recipients' H&Y score (Table 6.4). Additionally, higher scores on factors 1, 2 and 3, which collectively represent psychological burden and impact on life partners' personal and social lives and where the item cross-loading occurred, were associated with higher scores on the Neuropsychiatric Inventory (NPI) and higher participant distress related to care recipients' neuropsychiatric symptoms. The longer duration of cognitive impairment correlated strongest with factors 2 (personal strain) and 3 (interference with personal life). Lower scores on the health-related quality of life (EQ-5D) were associated with factors 2 and 4, the former factor represents current health state and the latter portrays concerns about life partners' future health and well-being in regards to their ability to provide care. Factors 1 and 4 were inversely correlated

with the caring role satisfaction sub-scale on the FCR and with positive interaction on the DRS. Lower life partners' age and shorter relationship duration only correlated with factor 2, and weekly care provision hours only correlated with factors 3 and 4.

Table 6.4 Correlations between life partners' and people with PRD variables and ZBI factor scores (n = 127 life partners).

		Factors				
	ZBI total	**1**	**2**	**3**	**4**	**5**
Life partner characteristics (n = 127)						
Age	-0.17	-0.21*	-0.28**	-0.09	-0.04	-0.07
Care provision duration (years)	0.12	-0.02	0.11	0.22*	0.09	-0.06
Care provision (hours a week)	0.19*	0.07	0.13	0.35***	0.18*	-0.12
Relationship duration	-0.16	-0.18*	-0.29**	-0.11	-0.07	0.01
RSS	-0.72***	-0.63***	-0.62***	-0.61***	-0.55***	-0.29**
Rel.SS	0.88***	0.71***	0.77***	0.79***	0.64***	0.34***
HADS-anxiety	0.68***	0.54***	0.68***	0.50***	0.49***	0.32***
HADS-depression	0.70***	0.53***	0.69***	0.60***	0.50***	0.23**
SF-12-physical health	-0.02	0.05	-0.12	0.02	-0.06	0.06
SF-12-mental health	-0.68***	-0.47***	-0.63***	-0.50***	-0.40***	-0.29**
EQ5D-index	-0.23*	-0.10	-0.31***	-0.15	-0.27**	0.05
EQ5D-VAS	-0.27**	-0.19*	-0.35***	-0.19*	-0.24**	-0.05
FCR-satisfaction	-0.15	-0.27**	-0.01	-0.06	-0.20*	-0.04
FCR-resentment	0.76***	0.62***	0.63***	0.73***	0.58***	0.29**
FCR-anger	-0.05	-0.01	-0.10	-0.05	0.03	0.02
DRS-positive interaction	-0.27**	-0.27**	-0.15	-0.15	-0.21**	-0.14
DRS-negative strain	0.72***	0.72***	0.59***	0.58***	0.58***	0.32***
BRS	-0.53***	-0.40***	-0.50***	-0.38***	-0.34***	-0.31***
Person with PRD characteristics (n = 127)						
Age	-0.14	-0.20*	-0.19*	-0.05	0.01	-0.07
Motor symptom duration	0.14	0.18*	0.07	0.19*	0.02	-0.02
Cognitive impairment duration	0.25**	0.19*	0.27**	0.27**	0.18*	0.07
Route one sub-sample of people with PRD (n = 57)						
NPI total	0.46**	0.39**	0.40**	0.50***	0.16	0.23
NPI caregiver distress	0.46**	0.42**	0.39**	0.48***	0.18	0.29*
H&Y stage	0.52***	0.43**	0.42**	0.43**	0.37**	0.45**

Notes: *p < 0.05, ** p < 0.01, *** p < 0.001

Abbreviations: BRS – Brief Resilience Scale; DRS – Dyadic Relationship Scale; EQ-5D – EuroQoL index score or visual analogue scale (VAS); FCR – Family Caregiving Role scale; HADS – Hospital Anxiety and Depression Scale; H&Y – Hoehn and Yahr; NPI – Neuropsychiatric Inventory; Rel.SS – Relatives' Stress Scale; PRD – Parkinson's-related dementia; RSS – Relationship Satisfaction Scale; SF-12 – Short Form 12 Health Survey; ZBI – Zarit Burden Interview.

6.4.6 Multiple linear regression analyses with factors

Prior to undertaking a regression analysis with each factor as the dependent variable, the assumption tests were carried out (Appendix H). Five multiple linear regression analyses were conducted with each factor as the dependent variable (see Table 6.5). A significant regression equation was found to predict factor 1 'social and psychological constraints ($F(7,105) = 29.065$, $p < 0.001$, adjusted $R^2 = 0.637$), factor 2 'personal strain' ($F(7,108) = 35.310$, $p < 0.001$, adjusted $R^2 = 0.676$), factor 3 'interference with personal life' ($F(7,106) = 32.750$, $p < 0.001$, adjusted $R^2 = 0.663$) and factor 4 'concerns about future' ($F(7,107) = 14.830$, $p < 0.001$, adjusted $R^2 = 0.459$), but not factor 5 'guilt'. Stress was a significant predictor for factors 1, 2, 3 and 4. In addition, factor 1 was predicted by negative strain sub-scale, relationship satisfaction and anxiety, factor 2 by anxiety, factor 3 by resentment sub-scale and factor 4 by relationship satisfaction.

Table 6.5 Summary of multiple linear regression analyses for variables predicting factors 1 – 5 (n = 127 life partners).

	Factors																			
	1				**2**				**3**				**4**				**5**			
Variable	*B*	*SE B*	β	*t*	*B*	*SE B*	β	*t*	*B*	*SE B*	β	*t*	*B*	*SE B*	β	*t*	*B*	*SE B*	β	*t*
RSS	-0.10	0.04	-0.22	**-2.26***	-0.07	0.04	-0.18	-1.97	0.01	0.05	0.01	0.15	-0.07	0.03	-0.24	**-2.04***	-0.03	0.03	-0.13	-0.87
Rel.SS	0.13	0.05	0.32	**2.92****	0.13	0.04	0.36	**3.49****	0.22	0.05	0.51	**4.81*****	0.07	0.03	0.28	**2.12***	0.04	0.03	0.21	1.26
HADS-A	0.24	0.11	0.25	**2.32***	0.20	0.08	0.23	**2.36***	-0.09	0.11	-0.09	-0.87	0.06	0.08	0.10	0.81	0.14	0.08	0.28	1.75
HADS-D	-0.07	0.13	-0.06	-0.53	0.18	0.10	0.18	1.80	0.08	0.13	0.06	0.59	0.04	0.09	0.05	0.41	-0.17	0.09	-0.30	-1.81
SF-12 MH	0.07	0.04	0.17	1.89	-0.01	0.03	-0.02	-0.18	0.01	0.04	0.02	0.17	0.05	0.03	0.18	1.61	-0.01	0.03	-0.05	-0.34
FCR-R	0.12	0.43	0.03	0.27	-0.23	0.35	-0.06	-0.66	1.54	0.44	0.33	**3.47****	0.41	0.32	0.15	1.28	-0.05	0.32	-0.02	-0.15
DRS-strain	0.45	0.12	0.34	**3.75*****	0.07	0.10	0.06	0.73	0.15	0.12	0.11	1.24	0.10	0.09	0.12	1.13	0.08	0.09	0.13	0.91
Adjusted R^2				**0.637**				**0.676**				**0.663**				**0.459**				0.134
F-value				29.065***				35.310***				32.750***				14.830***				3.531**

Notes: *p < 0.05, ** p < 0.01, *** p < 0.001

Abbreviations: B – unstandardized beta; β – standardised beta; SE B – standard error of unstandardized beta; t – t-test statistic; DRS-strain – Dyadic Relationship Scale, negative strain sub-scale; FCR-R – Family Caregiving Role scale, resentment sub-scale; HADS-A – Hospital Anxiety and Depression Scale, anxiety sub-scale; HADS-D – Hospital Anxiety and Depression Scale, depression sub-scale; Rel.SS – Relatives' Stress Scale; RSS – Relationship Satisfaction Scale; SF-12 MH – Short Form 12 Health Survey, mental health sub-scale

6.5 Discussion

6.5.1 General discussion

This is the first exploratory factor analysis of the Zarit Burden Interview undertaken with life partners of people with Parkinson's-related dementia. Life partners in this study experienced similar amount of burden as care partners of people with dementia and were of similar age (Ankri et al., 2005; Smith et al., 2018). However, the perceived burden in partners of people with PRD was greater than in partners of people with PD-MCI which is consistent with existing evidence (Grün et al., 2016; Leroi et al., 2012a; Martinez-Martin et al., 2015; Mosley et al., 2017). The reasons why burden increases with the progression of PD and cognitive impairment include disease-related aspects (e.g. longer disease duration, functional dependency, more severe motor impairment and neuropsychiatric symptoms, loss of ability to perform daily activities) (Leroi et al., 2012a; Martinez-Martin et al., 2008, 2015; Mosley et al., 2017; Santos-Garcia & de la Fuente-Fernandez, 2015; Schrag et al., 2006) and life partner related aspects (e.g. own depression, anxiety, stress, relationship satisfaction, health-related quality of life and psychological well-being) (Martinez-Martin et al., 2008; Mosley et al., 2017; Schrag et al., 2006; Shin et al., 2012). In addition, the current study identified that lower resilience, and higher negative strain and feelings of resentment were contributors of burden. These findings highlight that both care recipient and care provider factors increase burden indicating a synergistic effect.

The present study revealed a five-factor model explaining 65.61% of the total variance which is higher than in other dementia studies (Ankri et al., 2005; Leggett et al., 2011; Smith et al., 2018; Springate & Tremont, 2014). Contrary to our initial hypothesis predicting a three-factor-solution, five factors emerged from the exploratory factor analysis suggesting that burden may differ in life partners of people with PRD in comparison to other types of dementia. This has also been evidenced by Shin et al. (2012) and Svendsboe et al. (2016) in comparative studies of care partner burden amongst PDD/DLB and non-PD type dementia.

The first three factors, which jointly described the mental and psychological burden of care provision on the lives of life partners, were closely related with some items loading onto more than one of these factors. Factors one 'social and psychological constraints', two 'personal strain' and five 'guilt' shared conceptual similarities with factors identified in earlier studies (Ankri et al., 2005; Cheah et al., 2012; Cheng et al., 2014; Leggett et al., 2011; Li et al., 2018; Oh & Kim, 2018; Pillemer et al., 2018; Siegert et al., 2010; Smith et al., 2018; Springate & Tremont, 2014; Tang et al., 2017; Torres et al., 2012).

Factors one and two described the psychosocial impact of providing care on the life partners' social and personal lives as their social relationships and personal health have suffered as a result of providing care to a person with PRD, resulting in an increase in negative feelings such as strain, stress, anger, frustration and embarrassment. In contrast to earlier research, the factors of 'frustration, embarrassment and/or anger' (Oh & Kim, 2018; Pillemer et al., 2018; Smith et al., 2018; Springate & Tremont, 2014; Torres et al., 2012; Unson, Flynn, Haymes, Sancho, & Glendon, 2016) and 'loss of control' (Cheah et al., 2012; Cheng et al., 2014; Li et al., 2018) did not emerge as separate factors in this study but were captured by the first factor instead. This could be explained by the existing interrelationship between wanting to socialise with friends and being unable to invite friends to visit due to worries about care recipients' behaviour and having lack of control over it. Furthermore, the diminished visits from friends and lack of opportunities for socialising could increase loneliness in life partners and result in higher burden; however, attending support groups could act as a protective factor against psychosocial burden (McRae et al., 2009).

The fifth factor portrayed participants' perceptions of their role as a life partner and feeling that the care they provided was insufficient. Despite the fact that factor five consisted of only two items, it is a common dimension in previous studies (Ankri et al., 2005; Cheng et al., 2014; Oh & Kim, 2018; Pillemer et al., 2018; Siegert et al., 2010; Springate & Tremont, 2014; Torres et al., 2012) and has also been named as

'worry about caregiving performance' (Cheah et al., 2012; Leggett et al., 2011; Li et al., 2018) implying that 'guilt' is an independently standing burden construct amongst life partners.

Factors three 'interference with personal life', which described the limitations that caring responsibilities have set on the lives of life partners, and four 'concerns about future', which depicted the feelings of fear and uncertainty regarding the ability to provide care, are relatively unique factors that emerged from the current study. The former factor has only been found in Torres and colleagues' (2012) study with care partners of people with obsessive-compulsive disorder, and the latter factor has only been found in Smith and colleagues' (2018) study with care partners of people with dementia. People with advancing PD and cognitive impairment can become more dependent on the life partner due to loss of functional abilities and competency in carrying out activities of daily living which can have a profound effect on the time and freedom of life partners (Vatter et al., 2018a). As a result, life partners can have less time for themselves and for interactions with others. Worry and concern about future (factor four) amongst life partners of people with PRD has been found qualitatively (Vatter et al., 2018a) as spouses expressed concern what would happen to the care recipients if they were unable to provide care. The fourth factor was associated with life partners' lower health-related quality of life which directly relates to their ability to provide care; this highlights the importance of life partner well-being throughout the disease trajectory of care recipients, enabling them to continue caring for their partner.

Stress was a significant predictor of the first three factors, which can be collectively observed as the psychosocial impact on life partners' personal and social lives and which mirror the definition of burden provided by Zarit *et al.* (1986) and George and Gwyther (1986), who worded that burden encompasses the physical, psychological, emotional, social and financial problems for life partners as a result of providing care. Stress and burden are strongly related (Santos-Garcia & de la Fuente-Fernandez, 2015) and used interchangeably on occasion (Svendsboe et al., 2016). Leggett et al. (2011) described that burden is key in the stress process as life

partners subjectively assess their role and situation as a care provider and choose between continuing their role or delegating the responsibilities to others. The first three factors were also associated with care recipients' higher neuropsychiatric symptom score and longer duration of cognitive impairment, indicating that non-motor symptoms impact on the personal and social life of the life partner and contribute to their psychological burden and negative feelings. Studies have demonstrated that both motor and non-motor symptoms of PD contributed to burden in life partners but cognitive and neuropsychiatric symptoms had a stronger effect (Aarsland et al., 1999; Leiknes et al., 2015; Mosley et al., 2017) which is supported by the findings of the current study.

In addition to stress, negative dyadic strain and resentment predicted factors one and three, respectively, which emerged as new findings from this work. On close inspection, the 'negative dyadic strain' and 'resentment' sub-scales were conceptually similar with factors 1 and 3 which may explain this relationship. Both of these sub-scales explored feelings of strain, anger, loss of control, visits from friends, presence/lack of other care providers and loss of own time due to care provision. The relatively low number of predictors could be due to the nature of the measures in this study which explored negative feelings and aspects of care provision rather than positive experiences, and Smith *et al.* (2018) concluded that seeing positive aspects of care provision can be protective against burden; thus, it is an important construct to include in future studies.

6.5.2 Methodological strengths

The current study applied an exploratory factor analysis technique to explore the dimensionality of the ZBI. Several authors have illustrated that the ZBI is multidimensional; however, the factor structure in a sample of caregiving life partners of people with PRD had not been previously undertaken. An EFA, which is used for scale development and determining the number of dimensions (or factors or latent variables) in a scale (Worthington & Whittaker, 2006), was a suitable method to examine the factor structure and determine whether similar factors

were extracted in this study compared to earlier studies of factor analyses of ZBI. An EFA is a dynamic approach which employs quantitative and qualitative approaches and allows flexibility in regards to choosing a specific method, factor rotation and extraction, determining the final factor solution and naming the factors (Worthington & Whittaker, 2006). As a result of running an EFA a simpler depiction of a scale is presented and the data represents significant patterns in the data which is beneficial in understanding latent variables such as burden.

Alternative methods to measure latent variables (aside from factor analysis) are latent class analysis, latent trait analysis and latent profile analysis. The use of an appropriate method is decided according to type of data, i.e. if the data consists of continuous latent variables then factor analysis is applied and if the data consists of categorical latent variable then latent class analysis is applied (Institute of Medicine of the National Academies, 2014). Both latent class analysis and factor analysis are used for data reduction and share some similarities, such as the latent factors (or classes) are derived from observed data and the decision regarding the number of latent classes is similar to that of determining the number of factors, i.e. the purpose is to find the highest explained variability with an appropriate number of factors/latent classes (Uebersax, 2009). However, the latent class analysis aims to group the data into clusters, groups, types of cases according to the multivariate categorical data (e.g. disease subtype, gender, groups of people with high/low burden) which should be independent from one another, which does not fit with the construct of 'burden' where variables are related and the data are continuous. The latent class analysis has been referred as a categorical-data equivalent to a factor analysis (Uebersax, 2009) but the two approaches have several differences. For example, the factor analysis is interested in the structure of the variables based on the exploration of the relationship between the variables (i.e. correlations) and the participants are qualitatively diverse along continua, whereas the latent class analysis is concerned with the structure of the cases or groups based on the latent structure and there are qualitative differences between the participants (Ruscio & Ruscio, 2008; Uebersax, 2009). In the factor analysis the factor loadings are regression coefficients and the factors are rotated, whereas in the latent class

analysis the classes are based on conditional item-response probabilities and rotation is not applied. Therefore, in line with the aims of this study to explore the factor structure of the scale and with the nature of the observed variables which are continuous, a factor analysis method was utilized in this study.

In the current study all items except one loaded onto five factors explaining a relatively high variability in comparison to other similar studies. All five factors had a good internal consistency suggesting the factors that had emerged had a good reliability and the items in the factors were closely related measuring the same concept or trait. The assumption tests and minimum sample size requirements (100 participants and at least five participants per item), as recommended by Gorsuch (1983) and Kline (1994), were met in this study which advised that the factor analysis was feasible to conduct. Recruiting a sample of life partners of people with PRD can often be challenging due to specificity of the inclusion criteria (i.e. diagnosed with PD and cognitive impairment or dementia, living together, in a partnership) and due to difficulties in reaching the population who may live in rural areas or be isolated (i.e. no access or choosing not to access the Internet or local charity groups such as Parkinson's UK). Therefore, recruiting above the target sample size was considered a strength in this study.

6.5.3 Limitations

A number of limitations should be acknowledged. The response rate of 43% in the current study is lower than in other postal questionnaire studies; for instance, Morley et al. (2013) and Jakobsen et al. (2011) had a response rate of 61% and 62%, respectively, and Laakonnen et al. (2008) a response rate of 77%. The lower response rate could be due to the questionnaire getting lost in the post, not reaching the potential participant or the researcher, having errors in participants' home address, not following up participants with a phone call, participants not receiving a monetary reward for taking part, not finding the time or energy to complete the survey or unwillingness to participate in a research study or disclose personal and emotional feelings. Edwards and colleagues (2002) noted that

questionnaires that were of interest to participants, provided monetary payment, were short, personalised, colourful and were sent by recorded or first class delivery postage increased the response rate alongside with communicating with potential participants prior to and after sending the questionnaires out and posting an additional copy of the survey to those that did not respond. A future study that recruits participants through a postal questionnaire study should consider following up participants via telephone call, sending out another copy of the questionnaire if they had not returned it, providing monetary incentives and using short personalised questionnaires with colourful ink.

The life partners in this study were recruited through the INVEST study (route one) and a postal questionnaire study (route two). In regards to the first phase, secondary data of participants from the INVEST study was used and their data were non-anonymous to enable the identification and data extraction of life partners and spouses for the purposes of this study. Including a selected number of participants who met the inclusion criteria indicated a non-random sample. In the second phase, a postal questionnaire was sent to those that screen-failed participation in the INVEST study which could be a potential source of bias in the sample as they had been already contacted by a researcher for the purposes of the INVEST study. However, as these participants were not sampled alone but formed part of the overall sample it was not considered a methodological issue. Furthermore, the data of motor and neuropsychiatric symptom severity of people with PRD could not be obtained in route two due to the nature of a postal questionnaire, which reduced the sample size for disease-related variables.

This study employed minimum sample recommendations (≥ 100 participants), which was considered appropriate for the purposes of this study, but 100 participants was considered a small sample size according to Comrey and Lee (1992), who stated that a sample size of 100 participants is poor, 200 is fair, 300 is good, 500 is very good and 1000 is excellent. Recently, a participant-to-item-ratio of 10:1 has been recommended (Morgado, Meireles, Neves, Amaral, & Ferreira, 2017), which is double of the current sample size. Increasing the number of participants

could potentially result in a more stable factor structure and the findings could be generalised to a wider population.

Factor analysis as a statistical method has several limitations. First, conducting a factor analysis is highly subjective and ambiguous in regards to flexibility of choosing a specific factor analysis and rotation method, determining the number of factors to be extracted, deciding what to do with items that cross-loaded onto multiple factors and interpreting the factors, which can be contradictive amongst researchers who are undertaking the analysis with the same measure (Roberson, Elliot, Chang & Hill, 2014). The factor solutions also depend on the variability of the questions in the scale, and whether they ask similar or different questions about the construct; this could not be changed as the EFA was done on an already developed scale. It can be difficult to select the number of factors due to varied recommendations by statisticians (Flom, 2017). Missing data, which occurred in less than 10% of the cases, should be approached with caution as having missing information can decrease statistical power and increase the risk of making conclusions that are inaccurate (Collins, Schafer & Kam, 2001; Morgado et al., 2017).

6.5.4 Future directions

A future study should employ a larger sample size with a 'participant to item' ratio of at least 10:1 (Morgado et al., 2017) and recruit more life partners of people with PD-MCI, PDD and DLB to facilitate comparisons between groups. The sample should include people from various cultural backgrounds and with different types of relationship (e.g. spouses, adult children, family members) to cross-validate findings in demographically diverse populations and increase generalisability to a wider population (external validity). Future research could also include a wider range of measures including person with PRD measures of motor, cognitive and neuropsychiatric symptoms and measures of positive aspects and experiences of care provision. Additionally, a longitudinal design could be applied to expand on the understanding of causal relationships between the variables, observe the model fit

between two or more time-points and provide in-depth information regarding construct, content and predictive validity.

6.6 Conclusions

The results in this work support and extend previous studies by providing a five-factor model of burden with 'interference with personal life' and 'concerns about future' emerging as unique factors from this study, which highlight that type of dementia can play a significant role in life partners' burden. The findings of this work suggest that providing information, education, training, and support interventions to life partners is important to mitigate stress, burden, negative feelings, emotions and experiences of care provision and help to maintain their well-being, quality of life and dyadic relationship.

CHAPTER 7: Experiences of female caregiving life partners of intimate relationships as cognition declines in Parkinson's disease: A qualitative study (Study 3)

This chapter has been published:

Vatter, S., McDonald, K. R., Stanmore, E., Clare, L., McCormick, S. A., & Leroi, I. (2018a). A qualitative study of female caregiving spouses' experiences of intimate relationships as cognition declines in Parkinson's disease. *Age and Ageing, 47(4)*, 604-610.

The current study is part of the INVEST project for which ethical approval was granted (Yorkshire & The Humber – Bradford Leeds REC 15/YH/0531). This chapter follows the consolidated criteria for reporting qualitative research (COREQ, Tong, Sainsbury & Craig, 2007) in order to maximise clarity of the method and results section.

7.1 Introduction

Cognitive impairment and dementia in Parkinson's disease (PD) are prevalent with up to 50% of people developing mild cognitive impairment (PD-MCI) (Goldman & Litvan, 2011) and up to 80% of people developing dementia (PDD) within 10 to 20 years of their PD diagnosis (Hely et al., 2008). Another common type of dementia is dementia with Lewy bodies (DLB) with a prevalence of 10-20% of all dementia cases (Mueller et al., 2017). PD-MCI is a well-known precursor to dementia (Emre et al., 2007; Hindle, 2010; Hobson & Meara, 2015; Janvin, Larsen, Aarsland, & Hugdahl, 2006) and both occurrence and severity of motor, neuropsychiatric and cognitive symptoms intensifies with the progression of cognitive impairment and dementia in PD (Leroi et al., 2012a), highlighting the complex needs of this population. It is well-evidenced that health and support care costs, as well as frequency and length of hospital stays, increases in PDD and DLB (Bostrom et al., 2007; Low et al., 2015; Mueller et al., 2017; Vossius et al., 2014) in comparison to cognitively intact PD but these costs are largely saved by the care provided by informal care partners, who

are usually spouses (Prince et al., 2014). Therefore, it is vital to focus on preserving care partnerships in order to limit and minimise the costs of healthcare.

The progressive and complex nature of PD increases the need for care, a role which is mostly fulfilled by a spouse or a life partner, but providing care can have an immense effect on the quality of life (Lawson et al., 2017; Martinez-Martin et al., 2005) and mental, emotional and physical well-being (Tan et al., 2012) of life partners and as a consequence increase burden, strain and stress (Martinez-Martin et al., 2015; Mosley et al., 2017). Due to the caregiving role and increased responsibilities, many life partners may neglect their own health and care needs and have been referred to as 'the hidden or invisible patients' in the literature (Brodaty & Donkin, 2009; Fengler & Goodrich, 1979; Ostwald, 1997). Several studies also suggest that a neurodegenerative condition in one partner can significantly affect long-term intimate relationships (Evans & Lee, 2014; Harris, Adams, Zubatsky, & White, 2011; Pozzebon et al., 2016). However, transitions in intimate relationships in the context of one partner developing Parkinson's-related dementia (PRD) have not yet been explored qualitatively. Thus, this study aimed to conduct qualitative interviews with life partners of people with PD-MCI, PDD and DLB in order to gain insight into the changes to the intimate relationships as a result of PRD.

For the purposes of this study, participants did not have to be married but could also be co-habiting with the care recipient. For clarity and consistency, participants who are co-habiting and either married or in long-term relationships will be referred to as life partners throughout this chapter.

7.2 Aim

The objective of this study was to explore changes in long-term intimate relationships in PD-MCI, PDD and DLB through the perspective of caregiving life partners. The overarching research question of this study was:

- How have intimate relationships change as a result of one partner developing PD-MCI, PDD or DLB?

7.3 Method

7.3.1 Design

The interviews followed a semi-structured interview format, which was deemed most appropriate for its flexibility to explore participants' views, beliefs and experiences. Semi-structured interviews are described as 'conversations with a purpose' (Holloway, 1997) allowing interviewees to express their opinions freely. Furthermore, this format of interview provides opportunities to build rapport with the interviewee by pausing (particularly when discussing a sensitive subject area), observing non-verbal body language, controlling the pace and depth of the interview and adapting the order of the questions accordingly.

7.3.1.1 Interview schedule

An interview schedule, developed prior to the interview (see Appendix I), was approved by the research ethics committee (reference number: 15/YH/0531) and used in all interviews. Members of the Patient and Public Involvement (PPI) group in the INVEST study reviewed the schedule prior to commencing the interviews. The schedule included open, non-leading and conversational questions about current and premorbid relationship satisfaction, various types of intimacies and the impact of Parkinson's disease and cognitive decline on the life partner. The questions regarding emotional, social, recreational, intellectual and physical intimacy were informed by the Personal Assessment of Intimacy in Relationships scale (PAIR; Schaefer & Olson, 1981). Following the first few interviews, additional questions were added to the interview schedule to reflect the areas of interest that had emerged from these interviews. Relevant probes and prompts were asked during interviews facilitating the in-depth exploration of a particular subject.

7.3.1.2 Visual analogue scale

On completion of the initial five interviews, an adaptation was made to the interview design by introducing a single quantitative measure asking participants to rate their current and premorbid relationship satisfaction on a horizontal visual analogue scale (Price, Bush, Long & Harkins, 1994). The scale ranged from 0 (very dissatisfied) to 10 (very satisfied) and appeared to be a clear and significant indicator of change in participants' relationship satisfaction (Appendix I). The values of the visual analogue scale are presented in Table 7.1 in the results section.

7.3.2 Sample

Participants were recruited to the INVEST study through memory and/or movement disorder clinics in four participating research sites in Greater Manchester and through local UK-based charity websites (e.g. Parkinson's UK, Join Dementia Research, Lewy Body Society). All participants in the INVEST study had signed an informed consent form agreeing to be approached for the interviews and participate in audio-recorded interviews at the time of consenting to the INVEST study (see Appendix C); therefore, only verbal consent was sought from the participants. A purposive selection of participants, in particular criterion sampling, was used to include participants whose partner had a diagnosis of PD-MCI, PDD or DLB to assure diversity of care recipients' diagnosis.

Participants were eligible to participate in the interviews if they met the following criteria: (a) currently providing care to a person with a diagnosis of PD-MCI, PDD or DLB, (b) being in a long-term intimate relationship, and (c) living together with the care recipient. It was estimated that twelve interviews would be sufficient to reach saturation within the sample as per the suggestions of Guest, Bunce and Johnson (2006), who stated that data saturation in a qualitative study is reached with between six and twelve participants.

7.3.3 Procedure

7.3.3.1 Pre-interviews

Majority (94.3%) of partners and life partners recruited to the INVEST study in four Greater Manchester sites were female participants; thus, potential participants invited to take part in the interviews were all women. In total thirteen life partners were approached via telephone and the study's procedure, aim, duration and location was explained. Permission to record the interviews with an audio-recorder was also confirmed with participants at this stage. Most life partners provided instant verbal consent to participate, whereas a few life partners needed time for consideration due to inability to leave the care recipient on their own for the duration of the interview. Twelve participants agreed to take part in the interview, and one participant refused due to lack of private space at home for the interview to take place and unwillingness to leave the care recipient unattended if the interview was to be undertaken outside of their home.

7.3.3.2 During interviews

Prior to commencing each interview, I explained the nature, purpose and procedure of the interview to the life partner. All respondents were informed that their participation in the interviews was voluntary and they could withdraw their data if they no longer wished to continue participating in the study. Participants were also made aware that they were allowed to have as many rest-breaks as they needed, to pause or stop the interview at any time, and to ask questions in case of any confusion or misunderstanding. Interviewees were not made aware of the researcher's personal goals and motives for undertaking the interviews to avoid the bias of participants.

All face-to-face individual interviews were conducted by one white female (S.V.), who had three years of qualitative research experience. Interviews were audio-recorded on a digital voice-recorder and field notes were made throughout

interviews to supplement the audio data gathered and make observations about any behaviour, body language and environmental aspects (Sutton, 2015). Eleven interviews were conducted at the participants' homes and one interview took place in a café due to lack of private space in the participant's home.

There was an existing rapport between the interviewer and interviewees as the researcher had met life partners as well as people with PRD through the INVEST study. Being acquainted with life partners was deemed beneficial because: (a) it allowed the creation of a safe and supportive environment for the interview to take place, and (b) the researcher had met the care recipients, therefore being aware of the person spoken about in the interviews. The researcher had experience working within neurodegenerative conditions and researching intimate relationships quantitatively.

7.3.3.3 Post-interviews

Following the interviews, the researcher kept a reflective diary to record observations, thoughts and reflections about the interviews. These notes can provide additional depth, richness and diversity to the interpretation of the interviews after transcription (Burgess, 1984). The digital audio files were transcribed verbatim in an anonymised manner, i.e. removing or rewording any identifiable information. The author transcribed seven out of twelve interviews, which allowed familiarisation with the raw data during transcription. Two interviews were transcribed by a research assistant working on the INVEST trial and three interviews were transcribed by a paid transcriber. Following the interviews, the opinions of life partners regarding feedback on transcripts and analysis results were not sought.

7.3.4 Analysis

Interviews lasted between 35 and 97 minutes. Verbatim transcripts were analysed in MS Office Word (Microsoft Corp.) using the inductive thematic analysis technique

whereby the codes and themes emerge from within the data (Braun & Clarke, 2006; Patton, 1990). Thematic analysis is considered to be the most suitable analysis for the purposes of this study as it allows flexibility, aims at understanding the data and provides rich, in-depth and complex data (Braun & Clarke, 2006). Thematic analysis also grants the direct portrayal of participants' personal views, opinions, beliefs, perceptions and experiences (Braun & Clarke, 2006). In an inductive approach the raw data is collected for the purposes of addressing research questions (data-driven), rather than pairing the findings with an already existing theoretical framework (deductive – theory-driven) (Braun & Clarke, 2006; Thomas, 2006). Therefore, an inductive thematic analysis was chosen for this work as it fitted with the aim of gathering views and experiences of life partners regarding changes in their long-term relationships.

Braun and Clarke (2006) recommend a six-step process of analysing qualitative data, which was followed in the current study. First, I became familiarised with the text by actively reading and re-reading the transcripts and starting the search for patterns and meanings. Then, by systematically working through the texts and paying full attention to each important and interesting item of the data, the generation of initial codes began. In this phase attention was paid to noticing any patterns, commonalities and nuances between and within data sets. Then, the codes were listed and the manual search for themes began to avoid the possibility of missing out any relevant and meaningful information and data. The codes were analysed and arranged into possible themes which were then reviewed and discussed with two other supervisors (K.R.M. and E.S.) to reach consensus and ensure clarity, coherence and brevity. Finally, these themes were written up in a logical, consistent, meaningful and concise way. Each theme was presented with example quotes that "capture the essence of the point the researcher is demonstrating" (Braun & Clarke, 2006, p. 93).

7.4 Results

7.4.1 Demographic details of participants

A total of twelve individuals participated in semi-structured interviews between November 2016 and March 2017 (see Table 7.1). All participants were white British females with a mean age of 69.3 years (SD = 4.8, range 63 – 78 years). Four care recipients had PD-MCI, five had PDD and three had DLB. For those with a diagnosis of PD, the average duration of PD was 8.1 years and the median PD severity score of Hoehn and Yahr was 2.0 (range 1.0 to 4.0). The majority of participants were married with the exception of one respondent who was co-habiting and the average duration of women's partnership was 44.8 years (SD = 9.8, range 20 to 57 years). Life partners had provided care for about 5.3 years (SD = 3.2, range 1 to 11 years) and devoted 125 hours per week to care provision duties (SD = 57.0, range 7 to 168 hours). The mean scores on the visual analogue scale for premorbid and current relationship satisfaction for life partners of people with PD-MCI were 8.8 (SD = 1.04) and 5.5 (SD = 3.28), respectively, and for life partners of people with PDD/DLB 8.8 (SD = 0.96) and 2.1 (SD = 1.65), respectively.

Table 7.1 Sample characteristics (n =12 life partners).

Participant ID	Participant age (years)	Partner's diagnosis	Partner's age (years)	Disease duration (years)	H&Y stage	Type of relationship	Relationship duration (years)	Duration of care provision (years)	Weekly care provision (hours)	Premorbid relationship satisfaction (VAS)	Current relationship satisfaction (VAS)
P01	64	PDD	67	18	2.0	Marriage	44	8	72	NR	NR
P02	78	PDD	77	8	4.0	Marriage	56	7	168	NR	NR
P03	65	PDD	68	12	2.0	Marriage*	20	10	168	NR	NR
P04	69	PD-MCI	77	5	2.0	Marriage	50	3.5	49	NR	NR
P05	63	DLB	64	7	3.0	Marriage	37	6	84	NR	NR
P06	67	DLB	76	6	2.0	Marriage	46	5	140	8	2
P07	75	PDD	77	3	2.0	Marriage	57	2	168	10	0
P08	73	PD-MCI	78	3	1.5	Marriage	50	1	7	8	5
P09	72	PD-MCI	74	3	1.5	Marriage	50	4	168	10	9
P10	72	PDD	74	17	3.0	Co-habitation*	40	11	140	9	2.5
P11	64	PD-MCI	67	4	1.0	Marriage	45	4	168	8.5	2.5
P12	69	DLB	73	10	2.5	Marriage	43	2	168	8	4

Notes: *Second long-term relationship indicated.
Abbreviations: DLB – Dementia with Lewy Bodies; H&Y – Hoehn & Yahr; NR – not reported; PDD – Parkinson's disease dementia; PD-MCI – Parkinson's disease and mild cognitive impairment; VAS – visual analogue scale.

A total of three themes and ten sub-themes emerged from the thematic analysis; details are presented in Tables 7.2 and 7.3, and Figure 7.1. The three themes that emerged were: (1) Altered relationship, (2) Care partner challenges, and (3) Acceptance and adjustment.

Table 7.2 Themes and sub-themes.

Themes	Sub-themes
1. Altered relationship	Emotional distance
	Role transition
	Communication
2. Care partner challenges	Responsibilities
	Negative repercussions
	Motor and non-motor manifestations
	Concerns about the future
3. Acceptance and adjustment	Marital contract
	Social support
	Resilience and coping

Table 7.3, the frequency of quotes table, illustrates the number of references made in regards to each of the themes and sub-themes in groups of life partners of people with PD-MCI, PDD and DLB. A total of 1040 references were made across all themes and disease groups. However, the uneven sample size in each group must be acknowledged and the following descriptions are provided as a guide in understanding what each of those groups considered important and therefore, should be approached with caution.

All life partners provided comments relevant to each of the sub-themes, demonstrating that their experiences are comparable regardless of the diagnosis of the person with PRD. Table 7.3 describes the amount of quotes life partners made in each of the themes: 'Care partner challenges' (51.1%) was most talked about with 531 references, followed by 'Altered relationship' (32.9%) and 'Acceptance and adjustment' (16%) with 257 and 167 references made, respectively. From the sub-themes, most emphasis was placed on 'Emotional distance' (257 quotes), 'Negative repercussions' (201 quotes) and 'Motor and non-motor manifestations' (195 quotes) across the three groups. Although the majority of the themes were proportionate in the three groups, the life partners of people with PDD and DLB

spoke more of the care recipients' cognitive impairment and decreased level of communication as well as increases in negative feelings and loss of own time, freedom and independence, which is also seen in Figure 7.1.

Figure 7.1 provides a visual overview of the ten sub-themes according to each clinical syndrome of the care recipient and demonstrates the similarities between the PDD and DLB groups despite the unequal sample size. Life partners of people with PD-MCI also experienced changes in their role, relationship and an increase in responsibilities and negative feelings; however, these were less prominent than in life partners of people with dementia. Finally, life partners in the DLB group exhibited stronger feelings of resilience, acceptance and adjustment than life partners of people with PD-MCI and PDD, as seen in the figure. The three themes and ten sub-themes are presented below with illustrative quotes.

Figure 7.1 Frequency of comments in sub-themes by clinical syndrome.

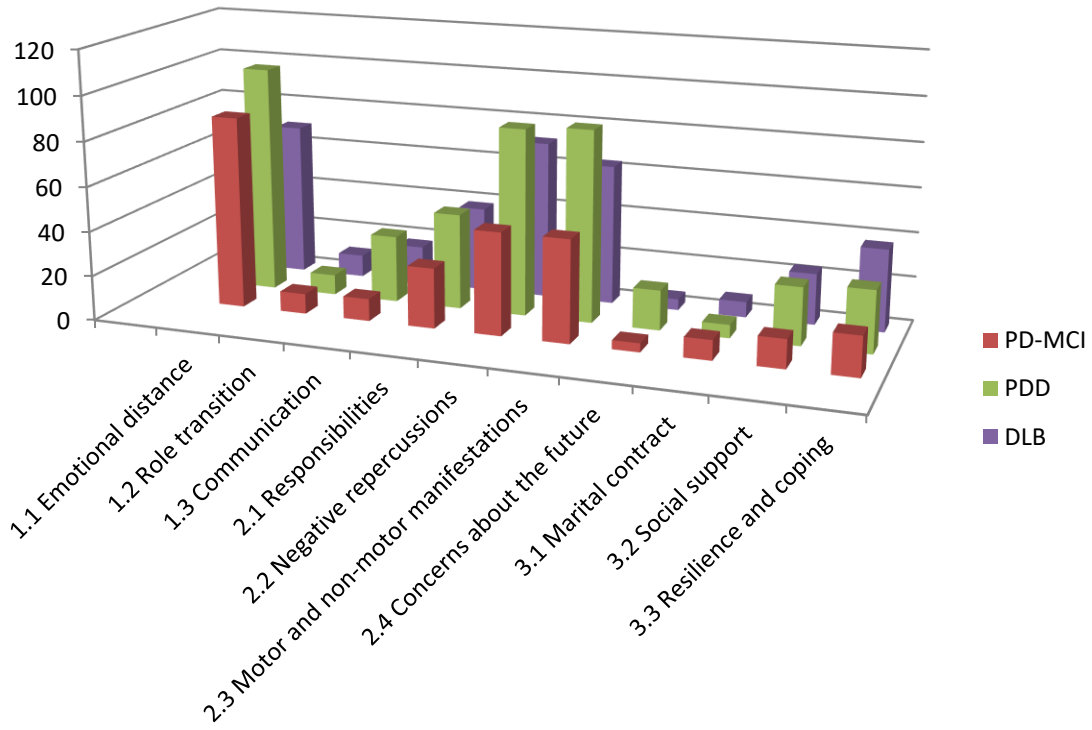


Table 7.3 Frequency of comments.

Life partner of people with: Themes, sub-themes and codes:	PD-MCI (n = 4)	PDD (n = 5)	DLB (n = 3)	Total (n = 12)
1. ALTERED RELATIONSHIP				
1.1 Emotional distance				
Premorbid relationship quality	13	11	6	
Altered relationship	11	14	7	
Altered intimacy	5	4	5	
Emotionally further & physically closer	14	15	12	
Emotional & intellectual intimacy	20	19	14	
Social & recreational intimacy	11	21	18	
Physical & sexual intimacy	10	13	4	
Loneliness	2	5	3	
Total of 1.1 Emotional distance	*86*	*102*	*69*	*257*
1.2 Role transition				
Own role changed	8	7	8	
Partner's role changed	1	2	2	
Total of 1.2 Role transition	*9*	*9*	*10*	*28*
1.3 Communication				
Decreased care recipient's level of communication	10	30	17	
Total of 1.3 Communication	*10*	*30*	*17*	*57*
Total of Theme 1 'Altered relationship'	***105***	***141***	***96***	***342***
2. CARE PARTNER CHALLENGES				
2.1 Responsibilities				
Own responsibilities increases	12	21	23	
Care recipient's loss of abilities & independency	15	22	15	
Total of 2.1 Responsibilities	*27*	*43*	*38*	*108*
2.2 Negative repercussions				
Own freedom, time, independency & life affected	3	17	15	
Care recipient's changed personality	20	15	12	
Resentment	9	11	6	
Burden	0	7	2	
Frustration & annoyance	8	10	17	
Sadness & grief	2	4	10	
Hopelessness & disappointment	0	4	2	
Worry & fear	3	11	4	
Guilt & distress	1	5	3	
Total of 2.2 Negative repercussions	*46*	*84*	*71*	*201*
2.3 Motor and non-motor manifestations				
Motor symptoms	12	28	15	
Cognitive impairment and dementia	17	39	31	
Neuropsychiatric symptoms	13	15	14	
Side effects from medication	4	4	3	

Total of 2.3 Motor and non-motor manifestations	*46*	*86*	*63*	*195*
2.4 Concerns about the future				
Deteriorating illness	3	10	3	
Inability to provide care & preparing for future	1	8	2	
Total of 2.4 Worry for future	*4*	*18*	*5*	*27*
Total of Theme 2 'Care partner challenges'	***123***	***231***	***177***	***531***
3. ACCEPTANCE & ADJUSTMENT				
3.1 Marital contract				
Commitment	6	5	5	
Reciprocal care	3	1	2	
Total of 3.1 Marital contract	*9*	*6*	*7*	*22*
3.2 Social support				
Friends and family	7	14	17	
Others (medical profession, respite or private care)	6	12	6	
Total of 3.2 Social support	*13*	*26*	*23*	*62*
3.3 Resilience and coping				
Resilience, coping & adjustment	12	17	23	
Acceptance	5	9	12	
Laughter	1	2	2	
Total of 3.3 Resilience and coping	*18*	*28*	*37*	*83*
Total of Theme 3 'Acceptance & adjustment'	***40***	***60***	***67***	***167***
Total number of comments for 3 themes	***268***	***432***	***340***	***1040***

7.4.2 Theme 1: Altered relationship

The theme 'Altered relationship' consisted of three sub-themes: 'Emotional distance', 'Role transition' and 'Communication'.

7.4.2.1 Emotional distance

The majority of participants (n = 10) noted in the interviews and on the visual analogue scale satisfaction with their marital relationship has declined as a result of their partners' neurodegenerative condition. However, participants whose partner had dementia had become more dissatisfied in comparison to those whose partner had mild cognitive impairment:

> *[We are] further, further, I mean I think if you ask [my husband] I think he would probably say closer because I know he relies on me for everything but I can't see it that way because it's not, it's not a closeness, it's a sympathy, you know, empathy. [P06, DLB]*

> *I don't think of us as a couple, no. [P03, PDD]*

> *There cannot be closeness when he doesn't know who you are. [P07, PDD]*

Global intimacy had decreased significantly to the point where it had become absent for some couples. Participants spoke of specific types of intimacies, such as emotional, recreational, intellectual, physical and sexual, and noted that all of them had become weaker in contrast to the premorbid stage. Women discussed that their partner was less able to understand their positive and negative feelings and they were less able to count on their partner in times of need (emotional intimacy). Life partners also reported that they did fewer social activities together and went out less than before (recreational intimacy) which lead to more time spent at home. In terms of intellectual intimacy, participants felt that the views and conversational topics that they shared with their partner had somewhat altered: some couples had more dialogues and discussions (e.g. about politics, television, past shared experiences) and some couples communicated less in comparison to the early stages of PD. Most life partners (n = 8) stated that they did not sleep in the same

bedroom as their partner due to symptoms related to PD (e.g. restless leg syndrome, night terrors, hallucinations, rapid eye movement sleep behaviour disorder) and noted that the level of physical closeness and intimacy with their partner, including hugs, holding hands, caresses, cuddles, and sex, had decreased:

> *Um, I don't think we have a relationship. Um, we live together as man and wife, but there is no sexual, there hasn't been for… 13 years. […] And there's no intimacy of any other sort. […] Um, he, he doesn't even hold my hand, you know, if you're walking, or put his arm around, there is nothing... nothing at all. [P11, PD-MCI]*

> *All of it [intimacy] has [changed], yeah. No physical intimacy. No social intimacy, he doesn't enjoy himself when he does go out. And emotional intimacy has never really been there [laughing]. P01, PDD]*

> *He knows he can't [get an erection] and he can't move anyway really. We used to laugh about that and say that we'd get a couple of hoists and we'd still have sex [laughs], with aids, um, um, but that was before the dementia. That was when we could chat about things like that and what about it, you know. [P07, PDD]*

> *I always give him a kiss when I leave him at night kind of thing, but in terms of the true couple intimacy then it has to be quite low. And that is because of the disease, because he takes up so much time and practical time, you know. [P06, DLB]*

> *Because of his illness we are doing less and less socially, so I find I'm in this house a lot of the time, you know, whereas we'd be out and about. [P12, DLB]*

Participants noted that due to the practical need of providing care and support to the partner and managing their partners' (instrumental) activities of daily living, for example making meals, driving, doing housework, managing medication and providing help with personal hygiene, they were spending more time together, but doing so had led to life partners feeling more distanced and separated from their partner at an emotional level:

> *We're together 24/7 and it's hard for everything to be all sweetness and light for 24 hours every day and you never get a break from it at all, you know. [P02, PDD]*

There isn't much scope for [relationship], because everything is related to caring, you know, it's cooking, dressing, washing. [P06, DLB]

As a result of the care recipients' diagnosis, some participants reported that the relationship had become uni-directional and the life partners felt lonelier and longed for connection, mutual companionship and reciprocity with their partners:

I have felt lonely when I'm with him. In fact, that's the time I do feel lonely, I don't feel lonely when I am on my own. [P03, PDD]

I just see myself very much as on my own but within a relationship where I can't do much because I'm not on my own. [P06, DLB]

7.4.2.2 Role transition

Life partners had different outlooks on their current role and were divided in their opinions. Four participants felt that their role as a wife had changed since the care recipients' diagnosis and they had become a caregiver. Half of the interviewees (n = 6) felt that their role had begun to transition and they alternated between the roles of a wife and a caregiver depending on the situation and care-related responsibilities and tasks. The role of a spouse remained unchanged for the two participants and they continued to see themselves as a wife. The views of life partners in terms of how they saw their role are presented below:

I don't consider myself a carer, no, he is quite independent. [P08, PD-MCI]

Well I say I'm his partner, but I care for him as well. [P10, PDD]

I've just got this person that needs looking after, I haven't got erm, a husband as such or a partner or a friend even, you know. […] I remember explaining this to somebody as it's like having somebody else's elderly uncle to stay. [P06, DLB]

Participants also noted that they began to see their partners in a different role, referring to them as a child and themselves as a mother looking after them:

It's like having a child, you are responsible for that child and watch them what they do and if you are and he is walking, "[partner's name], there's a step there, watch what you are doing'. [P10, PDD]

7.4.2.3 Communication

A significant change that all participants experienced was a diminished level of communication and fewer conversations with their partner which led to emotional distance from their partner. Although decreased communication was noted by all life partners, it had reduced more once dementia had emerged and become non-existent for several couples:

I miss the conversations, the natters, the chatters, the just saying, "Ooo did you enjoy that?" and talking about summat [something] you've watched, seen or done. […] We'll talk, but it's not a conversation. It's sort of "yes, no". [P11, PD-MCI]

You can't have a proper conversation, you might be saying something to him and then he'll answer you with something that's nothing to do with what you are talking about. [P10, PDD]

He cannot communicate. He cannot hold a sentence, he cannot say a sentence. […] He can't, you cannot discuss anything with him. It's… if, if we were recorded all day, it would just sound stupid rubbish, 'cause [my husband] just, when he can manage to say anything, it's just rubbish. [P07, PDD]

My children have commented that he's very quiet, he is going quieter as time goes by. [P12, DLB]

Two life partners acknowledged their husbands' decreased ability to communicate and accepted the change:

I am satisfied [with the level of communication] because it is all we can do out of a bad situation. [P06, DLB]

I talk to myself quite a lot while he is there, I'll read something and I'll say 'Oh that's really interesting. Oh look, such and such just happened' and there will be no response, but I think well that's alright [laughs]. [P03, PDD]

7.4.3 Theme 2: Care partner challenges

The sub-themes of the 'Care partner challenges' theme were: 'Responsibilities', 'Negative repercussions', 'Motor and non-motor manifestations' and 'Concerns about the future'.

7.4.3.1 Responsibilities

The progressive nature of the care recipients' neurodegenerative condition had brought about many changes for the couple and significantly increased responsibilities for the life partners who now had to integrate care provision tasks with their regular daily duties. Many people with dementia had lost the ability to drive, self-care, manage finances, do maintenance and administer medication, and therefore, wives had to take over their partners' previous obligations:

> *I've had to take on all the responsibility, money, power of attorney, I have to do the maintenance. [P01, PDD]*

> *You will look around and whatever you see, I do. Everything. I move the furniture, I, I cook, I, everything. He can't make a cup of tea, he can't switch the television on, he can't answer the phone, he can't clean himself up when he goes to the toilet. I do everything. […] When [my husband] isn't here, I spend all my time doing paperwork, phone calls, or, or well the decorating and things like at the moment, sorting the building stuff out. […] You don't have time to yourself. You're catching up then with the things that you can't do while you're [watching him]. [P07, PDD]*

With the emergence of dementia, care recipients' ability to complete (instrumental) activities of daily living had decreased and life partners felt that they had to become the wife and the husband in the relationship. Participants became increasingly responsible for their partner as well as his relationships with other people, contributing to the weight of responsibilities carried on their shoulders:

> *I do, I feel responsible for him. I'm not sure how much I should feel responsible for him because I think I take too much on myself. [P05, DLB]*

I do feel alone in the, because nobody, I won't say cares as much as I do, but is in, you know, charge of everything as much as I am or I can't, I know I can kind of get somebody else to take him places but I can't rid myself of the responsibility, it is just me. [P06, DLB]

People with PD-MCI had largely preserved their independence and ability to do things, as they could drive, self-care, take medication and manage finances and paperwork, but their life partners were beginning to check medication adherence and accuracy of completed paperwork:

He still likes to do the [finances], but I do find myself checking. [P08, PD-MCI]

7.4.3.2 Negative repercussions

The increase in spousal responsibilities had limited life partners' own time as a large proportion of the life partners' day was spent on providing care and surveillance, maintaining security and safety, and supporting their partner with daily tasks. As a result of providing continuous care, seven life partners whose partner had dementia felt that they had lost their own freedom, independence and life:

Sometimes in the morning I get in a panic 'cause I think I can't do this. […] I have no, no life and I have no future, I can't do anything. [P07, PDD]

I mean obviously everything's changed, it's got to. Sometimes I think I want me life back in that respect, you know. [P10, PDD]

I got to the point where I wasn't sleeping, I wasn't eating, I was crying, because I can't go out and leave him. I've got no freedom. And the doctors put me on some tablets for stress and they are helping. But it affects your whole life, all my freedom has been taken away from me. [P12, DLB]

Significant changes in life partners' own time and life were accompanied with a rise in negative feelings, such as frustration, sadness, grief, annoyance, disappointment, hopelessness, guilt, distress and worry:

It makes me sad because I know he can't help it. [P09, PD-MCI]

I feel frustrated at the moment and I feel a bit upset actually. Erm and I feel annoyed with myself of feeling upset because it's a vicious circle really, because things aren't how they were and I feel as though I ought to appreciate that more than I do. [P08, PD-MCI]

Grief has all these stages, you know. And I think with Parkinson's and dementia sometimes you are walking them stages while they are still alive. [P11, PD-MCI]

I want to run away sometimes [cries]. Um… I cope better with it now, 'cause I'm sort of getting a bit more used to it, but I feel sick [voice breaks with emotion], I feel resentment, I feel lost… It's just everything. [P07, PDD]

Resentment towards Parkinson's disease, cognitive impairment, the care recipient and the situation they were in was reported by several participants:

Sometimes you say to yourself "What did I do to deserve this?" Obviously it would be nice to go through life and not have an illness. [P01, PDD]

I do feel resentful sometimes when I think he's taking me for granted and I know I shouldn't do but I do […] because I know if it was me he wouldn't feel like that [cries]. [P02, PDD]

I'm not doing things that I want to do that I feel that I've earned the right to do after working all me [my] life and so some of that is resentment to him, some of it is resentment to the illness. And some of it is just resentment with life in general, you know because that's just how it is. [P05, DLB]

A few participants noted that their partners had become more self-absorbed and self-focussed as a result of their disease and became less attentive, apprehensive and observant about people around them. The changes in care recipients' personality also contributed to increased negative feelings, but one life partner recognised why this change occurred:

[He is] quite focused on the self but I understand it's like fighting for survival, you got to focus on yourself, just to get through the day if things are difficult, you know. But it's annoying as well [laughs]. [P03, PDD]

7.4.3.3 Motor and non-motor manifestations

Many times participants had separated non-motor symptoms, such as cognitive impairment and neuropsychiatric disturbances, from motor symptoms of the care recipients' illness and noted that the cognitive decline and neuropsychiatric symptoms, regardless of disease severity and duration, were significantly more difficult to accept, manage and cope with than the physical symptoms of Parkinson's disease. All life partners expressed that they were constantly involved in the management of their partners' motor and non-motor disease aspects, but challenges in handling with cognitive and neuropsychiatric symptoms were more pronounced in life partners whose partner had been diagnosed with PDD and DLB:

> *Dementia is very much more worrying. I mean with Parkinson's there's always the possibility of drug therapy to make that symptom better but there's nothing for dementia. [P01, PDD]*

> *With the Parkinson's we were still playing, um, Scrabble. We were still discussing, still sharing, yeah. All, all those things. Still loving. Not able to actually have a sex act, but still lots of loving. Lots of sharing. Uh, I could still, um, be upset and [my husband] would still put his arms around me and, and hug me and comfort me. But not now. But that's not the Parkinson's, that's the dementia. […] Once dementia's there, you're lost. If [my husband] was in a wheelchair I would cope admirably. If I had to bathe him, wash him, dress him whatever, whatever physically I would cope. […] I'm used to caring, you know, but not the dementia it's, it's evil. And there is no joy at all with dementia, there just isn't. [P07, PDD]*

> *I've coped with the Parkinson's fine but it's the dementia side of it which is the thing that gets me more than anything. […] If it was just Parkinson's we could carry on but the Lewy body is the main hurdle for us. [P12, DLB]*

7.4.3.4 Concerns about the future

Incessant care provision prompted participants to think of the future and they were worried what might happen to the care recipients if they were no longer able to provide care, for example due to deterioration of own mental and physical health or them dying before their partner. Many women also feared the inevitable progression of dementia leaving them incapable of planning for the future:

Well I suppose the most worrying thing for me is the future. […] I just wish [things] were how they were before, only because of not knowing what the future holds. [P08, PD-MCI]

I have to realise that it won't change. Because it's a progressive illness, it's not something that just stands still. [P09, PD-MCI]

I do worry about the future, I do worry that what, what will become of us in the future because I can't ever see us not being together but I worry what would happen if I went first. Who would look after him then? [P05, DLB]

I fear for the future, I do. I wonder how I'm going to cope, you know, if he gets really bad with it. Erm I mean, you know, I have to admit it is very difficult dealing with him physically you know because he's a big man. [P02, PDD]

Despite the challenges of making future plans, one wife described that she had asked her son to arrange care for her husband in case she was unable to provide care:

I've said to [my son] you know 'Dad wouldn't be able to look after himself if anything happens to me'. So I said 'If you've got to find somewhere for him to go, please look for the best that you can manage you know'. And he promised me he would. [P02, PDD]

A few participants described their anticipation for both partners to retire so that they could spend more time together with each other; however, the womens' dreams and plans for retirement had quickly vanished due to their partners' unexpected neurodegenerative diagnosis:

What a shame, because he had just finished work, because that was going to be a new thing for us really. [P06, DLB]

Once he was diagnosed with Lewy body I hand in my notice at work. [P12, DLB]

7.4.4 Theme 3: Acceptance and adjustment

The final emerging theme 'Acceptance and adjustment' was further divided into three sub-themes: 'Marital contract', 'Social support' and 'Resilience and coping'.

7.4.4.1 Marital contract

Providing care to one's partner was perceived as an integral part of the marriage agreement and life partners readily acknowledged that they would continue providing care and support to their partner in the future. Despite the challenges of care provision, life partners exhibited many positive feelings such as love, commitment, compassion, empathy, sympathy and altruism towards their spouse. Throughout the couple's marriage, people with PRD had often cared for their partners, who had been faced with challenging ailments, and wives felt it was their turn to look after their spouses:

> *He'd looked after me, so it's my turn now, I have to be the one for him. [P11, PD-MCI]*

> *When we got married you got married forever, you know. And that was it, for better, for worse and I always think you know, well you say in sickness and in health, well we've had the health bit and now we're on the sickness bit you know. It's just inevitable and you just have to accept it. […] And I've got to look after him because it's what I signed up to do all those years ago, you know [laughs]. [P02, PDD]*

7.4.4.2 Social support

Over half of couples (n = 8) were receiving informal social support from their adult children and other family members which they found helpful:

> *We have a very supportive family so we enjoy visiting family and they visit us and we have grandchildren. [P08, PD-MCI]*

> *My children always say to me "There's three of us looking after the dad, you are not on your own", so I find that very re-assuring, they are very good. [P12, DLB]*

Friends were considered another source of social support for the couple but participants acknowledged that due to the care recipients' deteriorating cognitive impairment and unpredictable behavioural patterns as well as friends' increasing age and health issues, the social interactions with their friends were becoming less frequent:

> *We've got, well we've got quite a lot of, well I won't say a lot but a handful of nice friends. They come and visit us, but at the same time a lot of our friends are now in the age group where they're all ailing with one thing or another, you know. [P02, PDD]*

> *There's a couple of friends [who] come, and we go for a short walk with them but people, I find, are not very understanding, they don't know how to deal with dementia. So rather than confronting they keep away erm. [P12, DLB]*

A third of life partners (n = 4) stated that they were receiving more help from the local voluntary sector groups (such as Parkinson's UK, Age UK) or church, which people with PRD attended either alone or with their spouse, than from their adult children and family members. Four couples were receiving support from formal paid carers or respite care. A few participants acknowledged that should they need (more) help, both formal and informal support was readily available:

> *It's getting into a routine where he is used to going [to respite] and during that day, that is my one day where I can either stay in and lie down as it were or I can do something without having to come back but that's only just been introduced. But that is supportive. [P06, DLB]*

> *I've not had a time where I've needed support […] but if I did need help then I would go to our children and they would [help]. [P08, PD-MCI]*

> *If I searched for the help I probably would get some [help] and I could pay privately for somebody to come in. [P01, PDD]*

7.4.4.3 Resilience and coping

The final theme referred to life partners' acceptance of the current situation and adjustment to it. Participants recognised that they were unable to change the

circumstances and instead focused on a variety of strategies, such as awareness, patience, acceptance, and coping, which helped them to move on with the situation. This was similar both in the case of mild cognitive impairment and dementia. A range of coping methods was reported by life partners and included laughter, separating the illness from the care recipient, recognising that the illness and care recipient's changing personality and behaviour was not of 'his making' and 'his fault' and applying previously learnt coping techniques to their lives:

> *One of the things that I was taught to do was to analyse myself every night, so I would say "What can I do about that?" Can't do anything about it, what's the point worrying about it. Cast it aside. And I do that you see. I am in a different position perhaps to a lot of wives, who've got husbands with Parkinson's because I have lots of methods of coping. [P09, PD-MCI]*

> *It's not something he can help, it's not something that he chose to have, it's not something that you know, it's not like he's been a drug taker and what's happened to him has, is a consequence of something he's done. It's not. It's just life, it's something that's happened and he can't change it and we have to live with it the best we can. So I certainly would never feel burdened with him. [P05, DLB]*

The majority of life partners exhibited great resilience and ability to accept and cope with the situation they were in and adjusting to it each day:

> *You just learn to adjust with what brings you every day 'cause it does, it's a degenerating disease and it brings you something new every day. You know, it's something else that you have to cope with. [P01, PDD]*

> *Sometimes I do feel a bit hopeless but I tend to bounce back again… [I] might wallow in self-pity [laughs] for a couple of hours or so and then think oh well you know, I get on with it each day you know. [P02, PDD]*

> *I am quite positive really with regards to the illness because to me you either fight it or you go down with it and both of us we'll go down with it, so you've got no alternative but to fight it. And to look at things we can do, not things we can't do, you can't dwell on the past. [P12, DLB]*

Finally, one participant treasured the fact that her partner was with her:

> *I just think that he's here and you know, my friend's husband's 63, passed away last week, had a massive heart attack, passed away and you know, you just think [my husband]'s getting up every morning, at least I've still got him here and no matter how difficult it gets, that's a bonus. [P05, DLB]*

7.5 Discussion

This qualitative study has provided important insights into the changes in long-term marital relationships as dementia progresses in Parkinson's disease. The analysis revealed three interlinked themes: changes in the marital relationship, challenges in providing care, and acceptance and adjustment of the situation, which are discussed below.

7.5.1 Altered relationship

The findings indicated that participants' relationship satisfaction had decreased as a result of their partners' neurodegenerative condition which was closely linked with partners' reduced ability to communicate and the transition in role for the life partners. Alongside reduced relationship satisfaction, global intimacy as well as emotional, social, recreational, intellectual, physical and sexual intimacies had altered and resulted in life partners feeling emotionally distanced from their partner despite spending more time together. These changes in intimate relationships in PRD resonate with qualitative research undertaken with life partners of people with dementia (Evans & Lee, 2014; Pozzebon et al., 2016), where transition and loss of relationship, partner, mutual companionship, reciprocity, connectedness, dyadic interaction and couplehood were frequent and common once one partner was diagnosed with dementia. Various terms have been proposed to reflect the changes that life partners are experiencing, for example 'husbandless-wives/wifeless-husbands' (Kaplan, 2001) and 'married-widows' (Baxter, Braithwaite, Golish, & Olson, 2002) reflecting the continuation of a relationship but without having the partner they once used to have.

The notion of being physically closer but feeling emotionally further away from their partner was recognised by most life partners in the interviews. This 'emotional disconnection' has been described in the field of dementia (Pozzebon et al., 2016); however, the 'physical closeness' due to day-to-day management of the condition was a finding that emerged in this study, which illustrated the unique challenges that PRD posed in this population. In the study undertaken by Baxter and colleagues (2002), they demonstrated that life partners whose husbands with dementia were either in a care home felt a conflict between their partners' physical presence and mental-emotional-cognitive absence, which presented several challenges and contradictions for the wives. On one hand life partners were keen to continue visiting their partners in a care home but on the other hand felt frustrated and sad by their husbands' worsening dementia and their inability to recognise their wives, which resulted in the removal of physical and emotional contact with their partner for some life partners (Baxter et al., 2002). The idea of the husbands' 'presence-absence' corresponded with the present work and the term 'married widowhood' (Baxter et al., 2002; Rollins, Waterman & Esmay, 1985) could also be applied to life partners of people with PDD and DLB.

The role transition was acknowledged by life partners in the current study and by life partners of people with dementia whereby they described their role as caregivers or at times as parents taking care of a child (Evans & Lee, 2014; Large & Slinger, 2015; Pozzebon et al., 2016). The change in one's role is personal and depends on several factors such as caregiving responsibilities, partners' abilities, independence and functionality, and own ability to cope, bounce back and adjust. Several spouses continued to view themselves as wives but with additional care-related responsibilities. One study (Molyneaux et al., 2011a) described how some life partners refused to be identified as 'carers' because they saw providing care as part of their relationship and marital commitment, whereas others endorsed their changed duties and responsibilities as that of the carer. Evans and Lee (2014) summarised that the altered couple's intimacy was directly associated with the dynamics in the relationship where one partner gradually became the care provider

and the other care recipient. Experiencing a change in one's role is directly linked to an increase in care provision responsibilities, emotional distancing from the partner and reduced relationship satisfaction as these processes happen simultaneously. The transition from a spousal role to care provider role is common in all types of dementia but some spouses maintain the dual role of a wife and a caregiver, which could be due to the length of the spousal role exceeding the length of the care provider role.

Communication and loss of ability to communicate in the person with PRD had contributed to relationship dissatisfaction and emotional distancing amongst all life partners in this study. Communication is regarded as one of the key elements in romantic relationships (Fitzpatrick & Best, 1979) and is directly linked to intimacy (Laurenceau et al., 2005; Mitchell et al., 2008; Yoo et al., 2014). Many studies in dementia have noted the loss of communication and its detrimental effect on relationships. Specifically, life partners of people with dementia found alterations in communication to be the most challenging aspect, which contributed to more negative feelings towards relationship quality and created a major disruption in the partnerships (Boylstein & Hayes, 2012; Clare et al., 2012; de Vugt et al., 2003; Egilstrod, Ravn, & Petersen, 2018; Evans & Lee, 2014; Pozzebon et al., 2016). This is understandable as lack of communication between the couple can act as a barrier to intimacy, closeness, reciprocity and bond. Care recipients' decreased ability to communicate, listen, focus and comprehend the meaning of the conversation was prominent once dementia had emerged and was often the central aspect around which the relationship satisfaction revolved. Life partners mentioned that even if they did speak to their spouses, it was not a meaningful dialogue, which many life partners missed. The concept of 'talking but not having a conversation' also appeared in the PD-MCI stage which is supported by Garand and colleagues (2007) who found that communication had reduced in people with MCI and this contributed to lower marital satisfaction in life partners. This suggests the importance of including people with varying stages of cognitive impairment in the study as their commonality of being on the dementia trajectory can present with

similarities in patterns such as decreased communication and reduced relationship satisfaction, regardless of disease stage.

Overall, the life partners of people with PRD had become emotionally distanced from their partners, started to experience a change in their role and noted reduced communication as one of the key elements in disrupting their satisfaction with the relationship.

7.5.2 Care partner challenges

Life partners shed light on the complex nature of the motor and non-motor symptoms of PRD as care recipients had lost skills and abilities to do things they were once capable of doing, which in turn increased life partners' responsibilities. Some life partners described they had a dual role in the marriage by being both the man and the woman in the relationship and managing the household, finances, maintenance, car which used to be their spouses' duty. These findings are consistent with previous studies with life partners of people with dementia where life partners took on additional responsibilities while providing care to their partners (Baxter et al., 2002; Boylstein & Hayes, 2012; Evans & Lee, 2014; Massimo, Evans & Benner, 2013; Pozzebon et al., 2016; Rollins et al., 1985). Evans and Lee (2014) described the changes in the marital relationships as a result of dementia a difficult process for both partners as one partner stepped back from their everyday duties that defined their role in the marriage, whilst the other life partner took on the new and unknown responsibilities which transitioned their role more into that of the caregiver. Life partners had to learn new skills, re-adjust their current commitments, make time for new responsibilities and find equilibrium between personal and care-related duties and tasks, which was closely linked with the role transition from wife to care provider. These changes were pertinent to life partners of people with PDD and DLB rather than PD-MCI as the independence and skills of people with PD-MCI was largely maintained.

The increase of care-related responsibilities due to the care recipients' condition was accompanied with an increase in negative feelings and took its toll on life partners. In particular, the time, freedom and independence of wives had reduced to the point of 'losing own life' and becoming mentally and physically weary. As a result of regular care provision, support and surveillance to their partners, wives felt a myriad of feelings such as resentment, frustration, annoyance, sadness, grief, despair, disappointment, guilt, distress and worry. The reasons for these negative repercussions varied from feeling burdened and stressed, not resting and sleeping, not comprehending care recipients' behaviour, having reduced interactions and reciprocity with partner, managing numerous care-related and household tasks, not finding the time to do things, having to postpone or cancel other commitments and having to stay indoors more frequently. Feelings of guilt, hopelessness, sadness, frustration, resentment but also loneliness and isolation have been reported in earlier studies (Baxter et al., 2002; Boylstein & Hayes, 2012; Pozzebon et al., 2016) and are frequently occurring in life partners of people with various types of dementia. Boylstein and Hayes (2012, p. 592) described the negative feelings of participants as 'disruption of marital closeness' and feelings of love, affection and acceptance towards their life partner 'as reconstruction of marital closeness'. The day-to-day experience of life partners providing care to their partners with PRD is closely related to how and what they feel, and what their commitments and responsibilities are, which in turn is associated with relationship satisfaction.

An important finding which emerged from this study was the manifestations of care recipients' motor, neuropsychiatric and cognitive symptoms, which were unique in PD-MCI, PDD and DLB and posed unique challenges for the life partners. Participants tended to separate cognitive impairment from neuropsychiatric and motor disturbances and perceived them to be harder to manage and put up with than the physical symptoms of PD. The frequency of quotes table demonstrated that in PD-MCI the motor, cognitive and neuropsychiatric symptoms were discussed in similar regularity by life partners, whereas in PDD and DLB non-motor symptoms, in particular cognitive impairment, received the most quotes and was described as much more worrying and difficult to cope with by life partners than motor

symptoms. This finding resonate with the quantitative studies where cognitive impairment and neuropsychiatric disturbances were found to be contribute to higher caregiver burden, distress and negative feelings in the care provider (Aarsland et al., 1999a; Martinez-Martin et al., 2015; Oh, Lee, Lee & Kim, 2015). Other quantitative studies also support the notion that care providers of people with PDD feel more burdened and have poorer quality of life compared to those providing care to individuals with cognitively intact PD or PD-MCI (Jones et al., 2017; Lawson et al., 2017; Leroi et al., 2012; Roland & Chappell, 2017). The difference between PD, PD-MCI and PDD highlights that the partners' progression of cognitive impairment and life partners' decrease in quality of life run parallel and may also be associated with higher relationship dissatisfaction for life partners of people with PD and dementia. The connection between care recipients' symptoms, care-related responsibilities, negative feelings and reduced relationship satisfaction was described by some life partners as a vicious circle. When care recipients' disease symptoms progressed, they became less independent and responsible, which led to an increase in spousal duties and tasks. Concurrently with the role transition in the couples' partnership, life partners experienced a variety of negative feelings, which in turn contributed to role transition from life partner to caregiver, a reduction in the relationship satisfaction and eventually loss of own life, independence and freedom.

Worries and concerns of life partners about the future occurred across all disease groups but were most prevalent in the PDD group as per frequency of quotes. Wives were concerned about the possibility of them being unable to provide care to their partners, for example due to deterioration of their own health or sudden death. These worries evoked life partners to think what might happen to their partners and who will take care of them, which consequently led some wives to make future arrangements of care provision for their spouses. Boylstein and Hayes (2012) summarised the various future prospects of caregiving life partners of people with AD as disruption and reconstruction of spousal closeness. Specifically, they found that 'holding on, acceptance, hope, breaking points, no future, and death' (Boylstein & Hayes, 2012, p. 592) were the most prominent recurring themes

about the possibilities that can happen in the future which also echoed with the life partners' perspectives in the current work. Life partners of people with dementia began to look towards the future, acknowledge the inescapable loss of their life partner and prepare themselves to face the future without their partner (Evans & Lee, 2014). However, the participants' concern about their life ending before the care recipients' life was a finding that was particularly marked in this study but not an apparent theme in other studies. This could potentially be due to life partners of people with PRD experiencing more burden, stress, worry and negative feelings in comparison to life partners of people with other neurodegenerative conditions as evidenced by quantitative studies, as well as the unique nature of PD-MCI, PDD and DLB that presents intricate challenges for the care partner.

The present study illustrated that relationship satisfaction had altered both at the mild and advanced stages of cognitive impairment but dementia had a stronger impact on life partners' lives, well-being and relationship satisfaction. This goes hand in hand with the length of the disease as life partners of people with PDD and DLB had provided care for longer than those providing care to partners with PD-MCI which can significantly contribute to increased negative feelings, burden, stress, and reduced relationship satisfaction. Participants had cared for their partners for over 5 years and spent 125 hours each week on care-related commitments and responsibilities. The former is comparable to the carers of people with dementia as 65.2% of carers had been providing care for over 5 years (NHS Digital, 2017). However, the latter is significantly higher amongst the life partners in this study in contrast to carers in the national adult carers survey (NHS Digital, 2017) as approximately 36% of carers spent more than 100 hours per week providing care to a person with dementia. In conclusion, the theme 'Care partner challenges' described life partners having to step up to new responsibilities, managing motor and non-motor symptoms of the care recipients' condition, experiencing a myriad of negative feelings and worrying about the future of their partners.

7.5.3 Acceptance & adjustment

The life partners accepted care provision as part of their marital contract and saw it inseparable from their commitments to the relationship with their partner. In spite of the challenges, difficulties and negative feelings that wives experienced and confronted with due to providing care, they revealed feelings of love, compassion, empathy and sympathy towards their partner. People with PRD had frequently cared for their wives throughout their married life when they needed help due to health ailments and this reciprocity was acknowledged by life partners who felt they ought to return the favour and care for their spouses until they can. Acceptance, adaptation, adjustment and preservation of a connection with the partner were important for life partners of people with dementia and seen as a positive strategy for maintaining the partnership in the present day (Pozzebon et al., 2016). Life partners in this study and in other qualitative studies were committed to their marital vows and held onto the 'in sickness and in health, til death do us apart' but there was also some confusion whether the marriage still existed as dementia progressed (Clark, Prescott & Murphy, 2017; Evans & Lee, 2014; Kaplan, 2001; Sanders & Power, 2009). Notwithstanding the conflict between existence and loss of relationship, life partners felt committed to their partners and were willing to continue providing care to their spouse in the future.

Several couples received social support from adult children, family members and friends which they found helpful and this is similar to previous research in dementia (Boylstein & Hayes, 2012). However, few life partners felt that friends did not visit as often as they used to due to the unpredictable nature of dementia and being unprepared to deal with it which reduced the amount of social interactions that couples had outside of their homes. Boylstein and Hayes (2012) found that although some friends were willing to provide help, many care partners did not accept the support provided by friends due to feelings of embarrassment and sadness about care recipients' behaviour. In the current study, participants received more support from adult children than friends but when informal help and support from family members was not available, couples received help from formal paid

carers and attending local voluntary sector groups. Receiving social support is important as it is associated with lower subjective burden (del-Pino-Casado, Frias-Osuna, Palomino-Moral, Ruzafa-Martinez, & Ramos-Morcillo, 2018) but informal support has a more profound effect on care partner burden than formal support (Shiba, Kondo, & Kondo, 2016), highlighting the importance of receiving help, support and assistance from friends and family. In contrast, when couples do not receive support, it can contribute to significant changes in intimate relationships (Boylstein & Hayes, 2012).

The final sub-theme described life partners' ability to accept, adapt and cope with the situation they were in. Life partners acknowledged their partners' neurodegenerative condition and their inability to alter the circumstances; thus, they learnt how to accept the condition and adjust to it daily. Many life partners had applied specific coping strategies, such as laughter, separating the illness from the care recipient, and recognising that the condition was not care recipients' fault. Earlier studies support these findings as life partners of people with dementia have learnt to adapt and adjust to the circumstances, use humour, separate the care recipient from the illness and focus on care recipients' needs, which helped life partners to move on and maintain their identity as a couple (Baxter et al., 2002; Boylstein & Hayes, 2012; Egilstrod et al., 2018; Pozzebon et al., 2016). Several life partners exhibited resilience as they were able to bounce back in a stressful situation, which resonates with earlier research (Pozzebon et al., 2016). It is important to focus on increasing and strengthening resilience as it can support care partners of people with dementia and help care partners to cope better with daily challenges (Parkinson et al., 2017). Additionally, accepting care recipients' diagnosis of dementia and the changes associated with the condition are paramount as couples can then receive access to support faster, which can subsequently help to preserve dyadic relationships (Singleton, Mukadam, Livingston, & Sommerlad, 2017).

7.5.4 Methodological strengths

Qualitative methodology collects detailed, in-depth information in an intricate manner. Participants can share their thoughts, feelings, experiences, emotions and perceptions in their own words, which does not segregate their responses into specific categories as quantitative research does (Anderson, 2010). Qualitative interviews are individualised allowing flexibility in terms of time, pauses, breaks, adjustment of the planned interview topic and exploration of additional questions that may have emerged in an interview. The data gathered in qualitative research contains personal perspectives and can offer a more dynamic, influential and vigorous understanding of a specific topic of interest (Anderson, 2010) and is considered helpful and valuable in the exploration of the question 'how'. Furthermore, the face-to-face contact between the researcher and the respondent in interviews provides the interviewer with an opportunity to make observations of the interviewee's use of words and body language which can be useful in finding out more about a specific topic area.

To my knowledge, interviews with life partners of people with PD-MCI, PDD and DLB regarding their long-term relationships and changes they might have experienced as a result of their partners' neurodegenerative condition have not been carried out. Furthermore, the current work collectively explored the combination of motor, neuropsychiatric and cognitive symptoms and looked at different stages of the cognitive decline in Parkinson's disease, which has been undertaken in one other qualitative study (Lawson et al., 2018).

An advantage of the current work is the inclusion of life partners of people with PD-MCI alongside dementia with PD as previous qualitative research has often focused more on dementia than MCI. Recently, qualitative studies with people with MCI and/or their life partners have grown in number (e.g. Carlozzi et al., 2018; Garand et al., 2007; Gomersall et al., 2015; Lu & Haase, 2009) adding to a better understanding of the changes both people with MCI and their life partners may go through. The interviews in this work provided an insight into life partners of people

with various cognitive stages in PD and demonstrated that life partners of people with PD-MCI also experience emotional distancing, decreased relationship satisfaction and intimacy, as well as increase in negative feelings, but to a lesser extent than in dementia. Including PD-MCI as a disease entity either separately or alongside with PDD and DLB is novel and expands on the knowledge of the impact that Parkinson's disease and either mild, moderate or advanced cognitive impairment has on life partners, their lives and relationship. Additionally, the interviewer's familiarisation with the interviewees' spouses through their participation in the INVEST study was considered beneficial as that permitted a more objective portrayal of people with PRD and their condition.

Including a quantitative element in qualitative research is less common but increasing in popularity. A frequency of quotes table was produced to ensure the inclusion of all relevant extracts in coding, themes and sub-themes from participants' interviews, to prevent unintentional elimination of any quotes by participants, and to provide a summary of the most quoted topic areas that life partners considered important. Current analyses demonstrated that three sub-themes emerged as the most prominent areas for participants, which simply presenting quotations would not have been able to do. The frequency of quotes also makes it possible to visualise the distribution of quotes within the themes and sub-themes in three disease groups, which is considered a strength in mapping the process of change in PD-MCI, PDD and DLB.

7.5.5 Limitations

The limitations of this qualitative study should be acknowledged. The interviews were undertaken only with white female caregiving life partners and did not include male life partners or other types of relationships, such as parent-child, same-sex or short-term relationships, which limits our understanding of whether ethnicity, gender or the specific nature of relationships can influence the experiences in intimate relationships in PRD. Ethnicity and cultural nuances are important in researching long-term relationships due to differences in cultural traditions and

norms which may set specific expectations in regards to care provision of family members and relatives. On the other hand, including a homogenous sample of white female life partners could be considered a strength as it provides a focused insight into the experiences of female life partners of people with PRD. The recruitment of female life partners was not an inclusion criterion but rather a reflection of the female-male ratio in the INVEST study where at the time of undertaking the interviews the ratio between female life partners (study partners) and male participants (people with PRD) was 33:2. Therefore, only women were approached for the interviews in this study. This is comparable to the sex ratio of carers in the UK as described in the Carers UK summary document (2017) where 78% of carers were female and 22% were male, and to the Alzheimer's UK summary document (2015) which stated that 60-70% of all informal dementia caregivers were women.

The interviews were undertaken with twelve participants, which was considered a sufficient sample size for a qualitative study (Guest, Bunce, & Johnson, 2006). However, increasing the number of participants in each of the disease groups (PD-MCI, PDD and DLB) could provide a deeper understanding of the experiences of life partners, and more variability and diversity of personal feelings, thoughts and experiences. Similar studies have undertaken interviews between 1 and 92 life partners and approximately half of these studies had a sample size below twelve participants (Pozzebon et al., 2016). The interviews in the current study highlighted that the impact of PDD and DLB on the life partners and their relationship satisfaction was more substantial in comparison to the PD-MCI stage and the investigation of changes in prodromal stages of dementia and diagnosed dementia in PD could be beneficial in gaining a deeper understanding of progression, transition and transformation of advancing cognitive impairment in PD.

The cross-sectional nature of this study collected participant views at one time-point only, therefore longitudinal observations of the relationships and comparisons between current and early stages of PD relationship satisfaction could not be made. Longitudinal studies of couples' relationships can be beneficial in

observing satisfaction, dynamics and quality of the relationship over time as well as couples' coping with stressful events and resilience. By understanding the complexities and nuances of couples' relationships we can predict the quality of the relationship in the future and potentially provide tailored interventions and strategies to maintain the relationship.

Finally, the chosen analysis was driven by an inductive thematic analysis approach as it was considered to be appropriate in investigating life partners' experiences in the interviews. However, it is noteworthy that thematic analysis may have limitations in comparison to other forms of qualitative analyses as it does not interpret data beyond the quotations, such as the interpretative phenomenological analysis and grounded theory do, and does not analyse nuances of language use, such as the conversation or discourse analyses do (Braun & Clarke, 2006). Despite these disadvantages, thematic analysis was selected as an appropriate and suitable method in addressing the aims of the current work, granting flexibility and production of rich meaningful data (Braun & Clarke, 2006). Qualitative research is also open to bias and ambiguity as data in the current study was primarily analysed by one researcher; however, one way this was addressed in the current study was involving two other researchers in order to achieve consensus and ensure coherence and clarity of the data. Another way that the agreement and consistency of the data could have been achieved, recommended by COREQ guidelines (Tong et al., 2007), was to seek the views and comments of participants on the transcripts, analysis and findings. By doing so the validity of researchers' findings could have been increased, the views, experiences and perspectives of life partners presented with higher accuracy, and the bias of researchers' own agenda kept to minimum (Popay, Rogers & Williams, 1998; Tong et al., 2007).

7.5.6 Future directions

A number of suggestions for future work are recommended. First, a subsequent study could use a heterogeneous sample and diversify the interviewee group by including male and female gender, people with various ethnicities and in distinct

types of relationship to maximise the number of different views, perceptions and experiences of being in an intimate relationship with a partner diagnosed with PRD. Second, researchers could consider splitting life partners of people with PD into mild cognitive impairment and dementia groups and undertake studies separately with larger samples to explore the experiences of life partners in the two groups in depth. And third, future qualitative work could be conducted with the couples together as it can provide rich data allowing the exploration of the perceptions and experiences of both partners, a comparison between the partners' perspectives, a meaningful observational data about how couples interact via verbal and non-verbal communication and a fuller awareness and understanding of the changes that the couple is experiencing and enduring (Bjornholt & Farstad, 2012; Mellor, Slaymaker, & Cleland, 2013; Taylor & de Vocht, 2011).

7.6 Conclusions

The current study explored changes in the intimate relationships of female life partners whose spouse had a diagnosis of PD-MCI, PDD or DLB. Interviews revealed three themes: alterations in the marital relationship, challenges in providing care, and acceptance and adjustment to the situation. The findings indicated that advanced cognitive impairment was associated with larger dissatisfaction with the intimate relationship in comparison to the early stages of cognitive decline in PD. Life partners of people with PDD and DLB transitioned from spouse to caregiver and had feelings of resentment, frustration, sadness and worry for the future. As dementia emerged, life partners experienced a loss of their own freedom and independence. Despite the fact that cognitive impairment and neuropsychiatric symptoms were difficult to manage, life partners honoured their marital vows and exhibited acceptance, resilience and adjustment towards the situation they were in. This study provides valuable insight into the changing patterns of long-term intimate relationships in Parkinson's-related dementia.

CHAPTER 8: Mutual influences on mental health and relationship satisfaction: an exploratory dyadic analysis of couples in Parkinson's-related dementia (Study 4)

This chapter describes a cross-sectional study of couples where one partner has a diagnosis of mild cognitive impairment or dementia in Parkinson's disease or dementia with Lewy bodies (collectively referred to as Parkinson's-related dementia, PRD) and applies an actor-partner interdependence model (APIM, Cook & Kenny, 2005; Kenny, Kashy & Cook, 2006). The study is currently in preparation for submission to a peer-reviewed journal.

8.1 Introduction

Life partners are crucial in helping and supporting people with mild cognitive impairment (PD-MCI) or dementia in Parkinson's disease (PDD) and dementia with Lewy bodies (DLB) (collectively referred to as 'Parkinson's-related dementia', PRD) with daily activities. However, continuous care provision can significantly increase their anxiety and depression (Aarsland et al., 1999a; Martinez-Martin et al., 2008; Schrag et al., 2006), and reduce mental health (Peters et al., 2011), life satisfaction (Aarsland et al., 1999a) and quality of life (Lawson et al., 2017; Leroi et al., 2012a; Martinez-Martin et al., 2008). The mental health (Aarsland et al., 1999a; Peters et al., 2011) and quality of life (Martinez-Martin et al., 2008) of life partners of people with PD is lower compared to general population and up to half of life partners can experience clinically significant anxiety and depression (Mosley et al., 2017). The duration of care provision in years and proportion of hours devoted to caring each day can lead to poorer mental health in life partners (Peters et al., 2011), which increases with the progression of cognitive impairment in PD. Once PDD or DLB have been diagnosed, life partners' depression (Roland & Chappell, 2017) and tensions in the dyadic relationship increase (Davis et al., 2011) and quality of life drops (Lawson et al., 2017; Leroi et al., 2012a; Szeto et al., 2016) in comparison to life partners of people with non-PD type dementia. This suggests that life partners

of people with PRD may have poorer health outcomes compared to care partners of people with other types of dementia, which has been confirmed in a recent comparative study (Wu et al., 2018).

People with PD may develop a number of neuropsychiatric symptoms, such as depression, apathy and anxiety (Chaudhuri et al., 2006; Chaudhuri et al., 2011; Jankovic, 2008), which remain prevalent once cognitive impairment has emerged in PD. In fact, the same neuropsychiatric symptoms are also frequent in PD-MCI and PDD (Aarsland et al., 2007; Martinez-Martin et al., 2015; Monastero, Di Fiore, Ventimiglia, Camarda & Camarda, 2013) as well as DLB. However, in addition to apathy, anxiety and depression, hallucinations and delusions are also common neuropsychiatric manifestations in DLB (McKeith et al., 2017). This is important as depression, alongside with duration and severity of PD, male gender and older age, are associated with cognitive impairment in PD (Aarsland et al., 2010; Litvan et al., 2011). Although studies have shown that anxiety and depression among people with PD-MCI can affect their quality of life (Schrag et al., 2000; Wiesli, Meyer, Fuhr, & Gschwandtner, 2017), less is known how these constructs are associated among people with PDD and DLB.

Earlier studies highlight that PD has a substantial impact on the couple's relationship but the effect can be greater once cognition has started to decline in PD. In research conducted within other degenerative conditions, relationship dissatisfaction among life partners is more pronounced in cognitive impairment and dementia (Davies et al., 2010; Garand et al., 2007) and significantly changes as a result of the person's dementia, sometimes to the point of relationship loss and termination (Quinn et al., 2009). In PD, it is well evidenced that non-motor symptoms are the most prominent stressors on couples' relationships once cognitive impairment has emerged in PD (Karlstedt et al., 2017); however, previous studies have not explored changes in relationships among people with PD-MCI, PDD and DLB and their life partners collectively.

Intimate relationships are the most profound connections we form in our lives (Hendrick, 2004) and the quality of the relationship can affect partners' outcomes. Specifically, couples who report higher relationship quality have better well-being, communication, more happiness and higher relationship satisfaction (Spanier, 1979), which helps to keep a relationship together for longer (Rusbult et al., 1998). On the other hand, lower relationship satisfaction, one of the components of relationship quality (Fletcher, Simpson, & Thomas, 2000), leads to higher depression scores (Baumeister & Leary, 1995; Beach et al., 1985, 2003; Clare et al., 2012; Levenson et al., 1993; Proulx et al., 2007), higher levels of relationship instability and dissolution (Gottman & Levenson, 1992), more aggression, criticism and blame as well as lower levels of intimacy (Beach et al., 1985) among individuals. Similarly, when one partner has higher depression scores, the other partner reports lower relationship satisfaction (Basco et al., 1992) highlighting that relationship (dis)satisfaction is closely linked to one's own health and well-being.

Given the complex, multifaceted and interdependent nature between the associations of the outcomes of each partner, it is important to study the outcomes of each member of the dyad together. To do this, an Actor-Partner Interdependence Model (APIM, Cook & Kenny, 2005; Kenny et al., 2006) was employed with couples of people with PRD and life partners to explore how the outcomes of one person can affect their own outcomes (i.e. actor effects) as well as the outcomes of their partner (i.e. partner effects). APIM will be conducted using multilevel modelling (MLM), which was considered the most suitable method to explore actor and partner effects as it takes into account the interdependence among dyads whereby each person (Level 1) is nested within the dyad (Level 2) (Kenny et al., 2006). MLM also allows the analysis of small sample sizes and is considered to be the most popular form of analysis for APIM as it "estimates all the parameters of the model within a single equation" (Cook & Kenny, 2005, p. 105). APIM has gained popularity among researchers over the last two decades as the model permits to study both members of the dyad together at once (Ledermann & Kenny, 2017), unlike many other linear models that assume independence between the participants. To date, only one pilot study of APIM exists in PD (Mavandadi et

al., 2014) and to my knowledge, this study is the first to jointly explore the associations of health-related outcomes and relationship satisfaction among life partners and people with PRD using APIM.

8.2 Aims

The purpose of Study 4 was to examine the associations among depression, anxiety, quality of life and relationship satisfaction among dyads of people with PD-MCI, PDD or DLB, and their life partners. Specifically, this study aimed to explore the actor effects (i.e. life partner) and partner effects (i.e. person with PRD) taking into account the interaction between life partners and people with PRD. Several research questions were proposed, which were tested using the Actor-Partner Interdependence Model (APIM; Cook & Kenny, 2005; Kenny et al., 2006), controlling for covariates.

The research questions are:

- What are the associations between **depression** and **relationship satisfaction** among life partners and people with PRD?
- What are the associations between **anxiety** and **relationship satisfaction** among life partners and people with PRD?
- What are the associations between **quality of life** and **relationship satisfaction** among life partners and people with PRD?

8.3 Methods

Chapter 4 provides a detailed overview of the design, eligibility criteria, recruitment, procedure, data collection, outcome measures and ethics of Study 4. The following sections briefly summarise the methods and analyses relevant to the current study.

8.3.1 Research design

This was a cross-sectional questionnaire-based study with people with PD-MCI, PDD or DLB, and their life partners, conducted as part of the baseline assessments in the INVEST study. Full ethical approval for the study was granted by the Yorkshire & the Humber – Bradford Leeds Research Ethics Committee (reference number: 15/YH/0531) (see Chapter 4, Section 4.2.8, and Appendix A). Capacity was assessed for all participants during the screening and informed consent visit (see Chapter 4, section 4.2.3.1). All participants provided written informed consent to participation in the study.

8.3.2 Participant sample and recruitment

Couples were recruited to the INVEST study from seven NHS sites in England between 12/04/2016 and 31/07/2017 (see Chapter 4, Section 4.2.2.3). People with PRD were eligible to participate if they had a diagnosis of PD-MCI, PDD or DLB according to the diagnostic criteria (Emre et al, 2007; Litvan et al., 2012; McKeith et al., 2005), if they were in a relationship and lived together with their partner or spouse. Life partners were eligible if they were a partner or a spouse of a person diagnosed with PD-MCI, PDD or DLB, and were not diagnosed with dementia.

8.3.3 Procedure

All couples completed sociodemographic and clinical assessments during screening and baseline visits with a researcher as part of the baseline assessments in the INVEST study.

8.3.4 Measures

A battery of socio-demographic (i.e. age, gender, education, ethnicity) (described in detail in Chapter 4, Section 4.2.3.4) and clinical assessments (described in detail in Chapter 4, Section 4.2.4.1) was completed by couples, including:

- The Relationship Satisfaction Scale (RSS; Burns, 1983),
- The Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983),
- The EuroQoL-5D-3L visual analogue scale (EQ-VAS; The EuroQol group, 1990).

Additionally, the following measures completed either by life partners or people with PRD were included as covariates.

Life partners:

- **Burden**: The Zarit Burden Interview (ZBI; Zarit et al., 1980).

People with PRD:

- **Cognition**: the Montreal Cognitive Assessment (MoCA; Nasreddine et al., 2005),
- **PD severity**: the Hoehn & Yahr stage (H&Y; Hoehn & Yahr, 1967),
- **Functional ability**: the Schwab & England Activities of Daily Living scale (SE-ADL; Schwab & England, 1969),
- **Neuropsychiatric symptoms**: the Neuropsychiatric Inventory (NPI; Cummings et al., 1994).

8.3.5 Analyses

All statistical analyses were undertaken in IBM Statistical Package for Social Sciences software for Windows, version 23.0 (SPSS, IBM Corp, 2015). The statistical analyses included descriptive (mean, range, standard deviations, percentage) and inferential (correlation tests, group mean comparisons, multilevel modelling) statistics. Categorical variables are presented as percentages and continuous variables as means and standard deviations. In order to verify the appropriateness of parametric tests for inferential statistics, the following assumption tests were undertaken: (1) normal distribution of residuals (P-P plot), (2) linearity (scatterplot

matrix), (3) homoscedasticity (residual plots), (4) lack of multicollinearity (variance inflation factor, VIF), and (5) outliers (boxplots) (Appendix J).

The assumption of independence, which is a standard assumption in linear models, is not applicable in the current study as the participants are couples, who influence each other and are therefore interdependent (Cook & Kenny, 2005). Thus, since there are non-independent observations, the unit of the analysis is the dyad, rather than the individual (Cook & Kenny, 2005; Kenny, 1995), and a specific model needs to be applied, which is discussed in detail below. For all results, the significance level was set at $p < 0.05$.

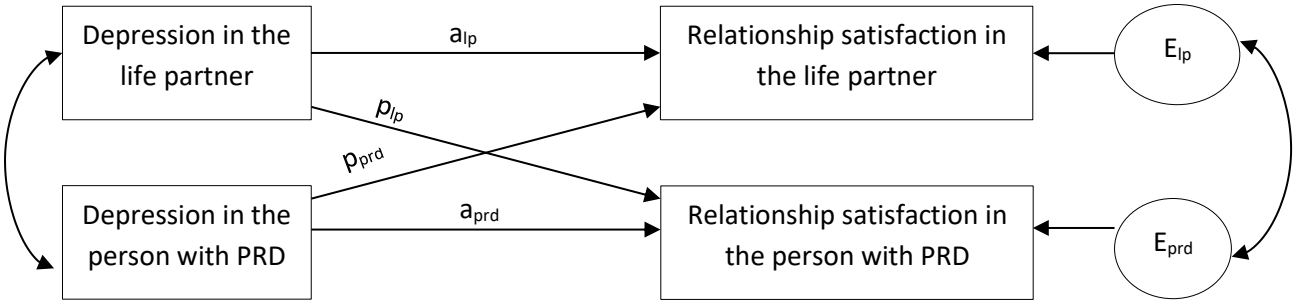
In order to verify whether values in the dataset were missing at random, Little's Missing Completely at Random (MCAR; Little, 1988) test was undertaken in SPSS. The Little's MCAR chi-square test result [(39, N = 114) = 35.539, p = 0.629] revealed that the data were missing at random, meeting the assumption for undertaking an imputation method to populate missing values. In the current study some missing information occurred in 1.8% of total values, in 5 variables (from a total of 10 variables) and in 11 participants from a sample of 114. On a variable level, missing data occurred between 0.9% and 5.3% (between 1 and 6 missing values per variable, respectively). The variables that had six missing values were RSS, HADS-anxiety and HADS-depression, which were all missing among people with PRD. There was a monotone pattern of missingness as assessed in the visual inspection of the missing value patterns matrix (Appendix J). Due to the robustness of multilevel modelling and the small percentage of missing data, the missing values were not imputed.

To address the research questions of the current study of examining the effects of life partner outcomes (i.e. actor) and person with PRD outcomes (i.e. partner) on the anxiety, depression, quality of life and relationship satisfaction of each partner, an Actor-Partner Interdependence Model (APIM) was conducted using multilevel modelling (MLM) (Cook & Kenny, 2005; Kenny et al., 2006). Prior to proceeding, the dataset was transformed into a pairwise dataset, whereby each row contained the

scores of the life partner and the scores of the person with PRD; this makes it possible to conduct dyadic data analysis in SPSS (Cook & Kenny, 2005; Kenny et al., 2006).

To account for the small sample size, a restricted maximum likelihood (REML) is generally recommended (Ledermann & Kenny, 2017) and was applied in this study. The current study was conducted with distinguishable dyads as the life partner and the person with PRD each had their own role in the relationship, which were coded as '-1' for life partners and '1' for people with PRD, as per Kenny *et al.*'s (2006) guidance. APIM assesses the *actor effects* (whilst controlling for partner effects) and the *partner effects* (whilst controlling for actor effects) (Cook & Kenny, 2005). The '*actor effects'* describe the degree to which the individual's (i.e. life partner) outcomes (i.e. relationship satisfaction) are predicted by their own scores (i.e. depression), whereas the *partner effects* are the effects of how much the partner's (i.e. person with PRD) outcomes (e.g. relationship satisfaction) are predicted by the scores of the life partner (i.e. depression) (see Figure 8.1).

Figure 8.1 An example of the actor-partner interdependence model in couples.



Abbreviations: a_{lp} – actor effect of life partner's depression on their relationship satisfaction; a_{prd} – actor effect of person with PRD's depression on their relationship satisfaction; E_{lp} – residual errors on relationship satisfaction for life partners; E_{prd} – residual errors on relationship satisfaction for people with PRD; p_{lp} – partner effect of life partners' depression on person with PRD's relationship satisfaction; p_{prd} – partner effect of person with PRD's depression on life partners' relationship satisfaction.

Six separate multilevel models were built to observe the associations among the studied variables. For all models, the subjects (i.e. dyad ID) and repeated value (the distinguishing factor, i.e. life partners or people with PRD) were entered and the

'compound symmetry: heterogeneous' option was selected which produced separate error variances for life partners and people with PRD (Kenny et al., 2006).

The models were as follows:

(1) the actors' and partners' depression scores (HADS) were the predictors and actors' relationship satisfaction (RSS) was the dependent variable;
(2) the actors' and partners' relationship satisfaction (RSS) were the predictors and actors' depression scores (HADS) was the dependent variable;
(3) the actors' and partners' anxiety (HADS) were the predictors and actors' relationship satisfaction (RSS) was the dependent variable;
(4) the actors' and partners' relationship satisfaction (RSS) were the predictors and actors' anxiety (HADS) was the dependent variable;
(5) the actors' and partners' quality of life (EQ-VAS) were the predictors and actors' relationship satisfaction (RSS) was the dependent variable;
(6) the actors' and partners' relationship satisfaction (RSS) were the predictors and actors' quality of life (EQ-VAS) was the dependent variable.

In all six models, the role of the person (i.e. the life partner or the person with PRD) was included as the distinguishing factor. All predictors were grand mean centred to avoid multicollinearity (Kenny et al., 2006). The models were first run without the covariates to observe unadjusted actor and partner effects. In the second step, the covariates (introduced as an interaction with the role) were entered one by one to observe whether any of the covariates impacted actor and partner effects between the independent variable and the dependent variable. The covariates were: relationship duration and age of both partners; life partners' care provision duration in years and weekly hours, life partners' burden score (ZBI); people with PRD's cognition score (MoCA), H&Y stage, SE-ADL score, NPI total score and NPI-apathy score. Only a few covariates (i.e. life partners' burden, NPI-apathy, and age) had a significant interaction effect with the studied variables and were included in the final trimmed models. The diagnosis of the person with PRD (i.e. PD-MCI, PDD, DLB) was not included as a covariate due to the small number of people with each clinical syndrome, which would significantly reduce the power of the analyses. The

gender of each member of the dyad was also not included as a covariate due to a large imbalance in gender (i.e. over 90% were male person with PRD-female life partner couples). Additionally, the results are presented without a correction and with a correction (i.e. Bonferroni post hoc test); however, as this study is exploratory, the uncorrected results will be interpreted.

8.4 Results

8.4.1 Characteristics of life partners and people with PRD

A total of 57 participant-life partner dyads participated in the study and their characteristics are provided in Table 8.1. All couples lived together and 53 couples (93%) were married. The average relationship duration was 45.4 years (SD = 12.80; range 10-61 years). The majority of participants were white British and over 90% were couples where the male had PRD and the life partner was female. The mean age of life partners was 69.5 years (SD = 6.94; range 48-85 years) and 74.0 years for people with PRD (SD = 6.64; range 55-90 years). Of the people with PRD, 18 had a diagnosis of PD-MCI, 25 of PDD and 14 of DLB.

According to the cut-off scores for each variable, 29 life partners (51.3%) and 16 people with PRD (31.4%) experienced relationship dissatisfaction (RSS cut-off ≤30), 6 life partners (10.5%) and 11 people with PRD (19.3%) had clinically significant anxiety scores, 5 life partners (8.8%) and 6 people with PRD (10.5%) had clinically significant depression scores (HADS cut-off ≥11), and 31 life partners (54.4%) and 41 people with PRD (71.9%) reported low quality of life (EQ-VAS cut-off ≤79).

Table 8.1 Participant characteristics (n = 57 participant-life partner dyads).

Categorical variables N (%)	**Life partners**	**People with PRD**	**t_{50}**
Gender, female	52 (91.2)	5 (8.8)	
Ethnicity, white British	52 (91.2)	53 (93.0)	
Relationship status, married	53 (93.0)		
Living with spouse/partner	57 (100)		
Retired, 'yes'	47 (82.5)	57 (100)	
Education			
Left school aged 14-16 years	21 (36.8)	26 (45.6)	
Left school aged 17-18 years	9 (15.8)	3 (5.3)	
Further education	14 (24.6)	15 (26.3)	
Higher education	9 (15.8)	6 (10.5)	
Postgraduate degree	4 (7.1)	7 (12.3)	
Diagnosis			
PD-MCI		18 (31.5)	
PDD		25 (43.8)	
DLB		14 (24.6)	
H&Y stage			
I		9 (15.8)	
II		28 (49.1)	
III		6 (10.5)	
IV		10 (17.5)	
V		2 (3.5)	
Continuous variables Mean (SD) [range]			
Age, years	69.5 (6.94) [48-85]	74.0 (6.64) [55-90]	
Relationship duration, years	45.4 (12.80) [10-61]		
Age left full-time education	17.2 (2.88) [14-31]	18.2 (5.31) [14-40]	
MoCA score (max 30)	27.9 (2.00) [22-30]	18.4 (5.39) [7-30]	
Duration of PD, years		8.4 (7.10) [1-37]	
Duration of cognitive impairment, years		3.9 (3.53) [0.2-16]	
Care provision duration, years	5.3 (4.64) [0-20]		
Care provision hours/week	98.8 (66.57) [0-168]		
RSS (max 42)	27.3 (11.47) [0-42]	32.9 (8.41) [12-42]	-3.60***
HADS-anxiety (max 21)	4.4 (4.07) [0-17]	6.7 (3.03) [0-13]	-3.51***
HADS-depression (max 21)	5.8 (4.19) [0-18]	7.2 (4.25) [0-17]	-1.85
EQ-5D VAS (max 100%)	75.0 (16.83) [35-100]	65.0 (17.03) [25-95]	2.99**

Abbreviations: DLB – Dementia with Lewy bodies; EQ-5D VAS – EuroQoL visual analogue scale; HADS – Hospital Anxiety and Depression Scale; H&Y – Hoehn and Yahr stage; MoCA – Montreal Cognitive Assessment; PD – Parkinson's disease; PDD – Parkinson's disease dementia; PD-MCI – Parkinson's disease and mild cognitive impairment; PRD – Parkinson's-related dementia; RSS – Relationship Satisfaction Scale; SD – standard deviation; t – t-test value; VAS – visual analogue scale.
Notes: ** $p < 0.01$, *** $p \leq 0.001$.

8.4.2 Outcome analyses

8.4.2.1 Assumptions for inferential tests

Before undertaking the statistical tests to observe the associations between life partner and person with PRD outcomes, and exploring the actor and partner effects, assumption tests were undertaken (Table 8.2; Appendix J). The probability-probability (P-P) plot revealed that the residuals were normally distributed, meeting the normality assumption. The assumption of linearity was met as all variables had a linear association. A visual inspection of the residual plots (standardised residuals versus standardised predicted values) showed that the data were scattered for all variables, suggesting that all variables had homoscedasticity. In terms of multicollinearity, a variance inflation factor (VIF) between 1.223 and 1.833 across all variables confirmed that there was no multicollinearity. Finally, a visual inspection of the boxplots revealed no outliers. As all the assumptions were met, the suitability of parametric tests was confirmed.

Table 8.2 Assumption tests of parametric tests for each outcome variable.

Measures / Assumptions	RSS	HADS-A	HADS-D	EQ-5D VAS
1. Normal distribution of residuals (P-P plot)	✓	✓	✓	✓
2. Linearity (scatterplot matrix)	✓	✓	✓	✓
3. Homoscedasticity (residual plots)	✓	✓	✓	✓
4. Lack of multicollinearity (VIF <10)	✓	✓	✓	✓
5. Outliers (boxplots)	0	0	0	0

Abbreviations: EQ-5D VAS – EuroQoL visual analogue scale; HADS – Hospital Anxiety and Depression Scale, anxiety or depression sub-scale; RSS – Relationship Satisfaction Scale; VIF – variance inflation factor.

8.4.2.2 Associations between the outcomes of partners

Pearson correlation analyses showed that higher relationship satisfaction was associated with lower depression, anxiety, and higher quality of life for both people with PRD and life partners (Table 8.3). Both partners were more satisfied with the relationship if they were older, had been in a relationship for longer and if the life partner experienced less burden. Life partners who were younger and who had

been in a relationship for a shorter time had higher burden scores. Additionally, people with PRD had more anxiety if the life partner had higher levels of burden and had provided more hours of care per week. The only outcome variable that correlated between members of the dyad was relationship satisfaction ($r = 0.422$, $p = 0.002$), which also confirms that the data were non-independent.

Paired t-tests were run to compare the outcomes between people with PRD and life partners, revealing statistical differences between the two groups (see Table 8.1). Specifically, life partners ($M = 27.63$, $SD = 11.29$) were less satisfied with the relationship than people with PRD [$M = 32.88$, $SD = 8.41$, $t(50) = -3.56$, $p = 0.001$]. Life partners also had lower anxiety scores ($M = 4.37$, $SD = 4.07$) compared to people with PRD [$M = 6.67$, $SD = 3.03$, $t(50) = -3.51$, $p = 0.001$], and higher quality of life score [$t(50) = 2.99$, $p = 0.004$] but there were no statistically significant differences in depression scores between people with PRD and life partners.

Table 8.3 Pearson correlations between outcomes of people with PRD and life partners (n = 57 couples).

	RSS	HADS-A	HADS-D	EQ-VAS	Age	Relationship duration	Care provision years	Care provision h/week	ZBI
RSS	**0.422****	-0.649***	-0.467***	0.278*	0.373**	0.268*	-0.012	-0.236	-0.668***
HADS-A	-0.409***	**0.164**	0.697***	-0.272*	-0.186	-0.282*	0.192	0.055	0.686***
HADS-D	-0.497***	0.532***	**0.237**	-0.317*	-0.205	-0.170	0.113	-0.120	0.688***
EQ-5D VAS	0.401**	-0.407**	-0.407**	**-0.099**	-0.048	0.001	0.051	-0.211	-0.177
Age	0.448**	-0.285*	-0.275	0.310*	**0.625*** **	0.628***	-0.161	-0.084	-0.329*
Relationship duration	0.316*	-0.185	-0.205	0.090	0.330*	N/A	-0.073	-0.032	-0.277*
Care provision years	-0.013	0.238	0.069	-0.093	0.079	-0.073	N/A	0.189	0.173
Care provision h/week	-0.142	0.340*	-0.026	-0.089	-0.138	-0.032	0.189	N/A	0.140
ZBI	-0.284*	0.331*	0.218	-0.089	-0.259	-0.277*	0.173	0.140	N/A

Notes: The correlations between life partners' outcomes are presented above the diagonal (shaded in grey); the correlations between people with PRD outcomes are presented below the diagonal; the bolded correlations between dyad members are presented along the diagonal. * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$
Abbreviations: EQ-5D VAS – EuroQoL visual analogue scale; HADS – Hospital Anxiety and Depression Scale, anxiety or depression sub-scale; RSS – Relationship Satisfaction Scale; ZBI – Zarit Burden Interview.

8.4.3 Actor-partner interdependence model (APIM)

Six separate multilevel models were run to observe actor and partner effects. In all six models, there were significant **actor effects** for life partners and people with PRD when the models were unadjusted for covariates (Table 8.4). Specifically, there were negative actor effects in models I – IV and positive actor effects in models V and VI. In models I and III, each person's higher depression or anxiety scores, respectively, predicted their own relationship dissatisfaction but not the relationship (dis)satisfaction of their partner. In models II and IV, each person's higher relationship dissatisfaction scores predicted their own depression or anxiety, respectively, but not the depression and anxiety scores of their partner. In model V each person's quality of life predicted their own relationship satisfaction and in model VI each person's relationship satisfaction predicted their own quality of life.

In the second step, each covariate was entered as an interaction term with the role (i.e. life partner or person with PRD; Table 8.5). A total of ten covariates were individually entered alongside the main actor and partner effects to observe the interaction effects. The covariates that had a non-significant interaction effect were excluded from the final trimmed model. After entering the covariates, all **actor effects** remained significant for people with PRD for the six models; however, for life partners, only models I, II, V and VI had significant **actor effects**. The directions of the effects were similar in models with and without the covariates, i.e. models I – IV had a negative actor effect and models V and VI had a positive actor effect.

In addition, two **partner effects** emerged for models III and IV. In model III, a higher anxiety score among people with PRD predicted a higher relationship satisfaction score in life partners ($B = 0.93$, $p = 0.004$). In model IV, relationship satisfaction of life partners predicted anxiety in people with PRD ($B = 0.14$, $p = 0.034$). These findings can be summarised in a feedback cycle (Figure 8.2). There is a positive bidirectional relationship between the anxiety scores of people with PRD and relationship satisfaction of life partners, wherein higher anxiety of people with PRD contributes to higher relationship satisfaction in life partners and higher

relationship satisfaction of life partners contributes to higher anxiety in people with PRD (Figure 8.3). Between the anxiety scores and relationship satisfaction of people with PRD, there is a negative bidirectional relationship as higher anxiety contributes to lower relationship satisfaction in people with PRD and lower relationship satisfaction contributes to higher anxiety in people with PRD (Figure 8.3).

8.4.3.1 Corrected APIM

Taking into consideration that several variables were explored, the models were adjusted with Bonferroni correction at alpha level of 0.0125 (0.05/4). With Bonferroni corrections, the actor effects remain significant for life partners in models I and II whereby depression scores and relationship satisfaction are correlated. For models III and IV, the actor effects are significant for people with PRD whereby higher anxiety contributes to lower relationship satisfaction and lower relationship satisfaction predicts higher anxiety in people with PRD. In addition, a partner effect exists in model III whereby higher anxiety scores of people with PRD contribute to higher relationship satisfaction in life partners. In models V and VI, no actor or partner effects are found between the quality of life and relationship satisfaction measures.

Table 8.4 Unadjusted actor-partner interdependence model between the associations of health outcomes and relationship satisfaction in couples of people with PRD and life partners (n = 57 couples).

	Life partners						People with Parkinson's-related dementia					
			95% CI						95% CI			
PREDICTORS	**B**	**SE**	**lower**	**upper**	**t-test**	**p-value**	**B**	**SE**	**lower**	**upper**	**t-test**	**p-value**
Model I: Depression	**DEPENDENT VARIABLE: RELATIONSHIP SATISFACTION**											
Actor effects	-1.77	0.31	-2.39	-1.15	-5.70	**0.000**	-1.04	0.39	-1.82	-0.26	-2.69	**0.010**
Partner effects	-0.31	0.42	-1.15	0.53	-0.74	0.466	-0.36	0.29	-0.93	0.22	-1.26	0.216
Model II: Relationship satisfaction	**DEPENDENT VARIABLE: DEPRESSION**											
Actor effects	-0.23	0.04	-0.32	-0.14	-5.14	**0.000**	-0.14	0.05	-0.24	-0.03	-2.54	**0.015**
Partner effects	0.02	0.06	-0.10	0.14	0.35	0.727	-0.01	0.04	-0.10	0.07	-0.37	0.715
Model III: Anxiety	**DEPENDENT VARIABLE: RELATIONSHIP SATISFACTION**											
Actor effects	-1.22	0.35	-1.93	-0.52	-3.50	**0.001**	-1.10	0.26	-4.18	-1.63	-0.57	**0.000**
Partner effects	-0.36	0.34	-1.06	0.33	-1.06	0.297	0.39	0.26	-1.34	0.92	1.50	0.141
Model IV: Relationship satisfaction	**DEPENDENT VARIABLE: ANXIETY**											
Actor effects	-0.22	0.05	-0.33	-0.12	-4.19	**0.000**	-0.24	0.07	-0.38	-0.09	-3.31	**0.002**
Partner effects	0.14	0.07	0.00	0.29	1.98	0.053	-0.03	0.05	-0.13	0.08	-0.47	0.638
Model V: Quality of life	**DEPENDENT VARIABLE: RELATIONSHIP SATISFACTION**											
Actor effects	0.20	0.09	0.02	0.38	2.28	**0.027**	0.20	0.07	0.07	0.33	3.12	**0.003**
Partner effects	0.11	0.09	-0.06	0.29	1.32	0.192	-0.01	0.06	-0.14	0.12	-0.10	0.919
Model VI: Relationship satisfaction	**DEPENDENT VARIABLE: QUALITY OF LIFE**											
Actor effects	0.52	0.22	0.07	0.97	2.33	**0.024**	0.87	0.29	0.28	1.46	2.97	**0.005**
Partner effects	-0.45	0.31	-1.06	1.68	-1.46	0.151	-0.11	0.21	-0.54	0.32	-0.50	0.622

Abbreviations: B – Beta; CI – confidence interval; SE – standard error.

Table 8.5 Adjusted actor-partner interdependence model between the associations of health outcomes and relationship satisfaction (n = 57 couples).

		Life partners						People with Parkinson's-related dementia					
				95% CI						95% CI			
PREDICTORS		**B**	**SE**	**lower**	**upper**	**t-test**	**p-value**	**B**	**SE**	**lower**	**upper**	**t-test**	**p-value**
Model I: Depression		**DEPENDENT VARIABLE: RELATIONSHIP SATISFACTION**											
	Actor	-1.06	0.35	-1.76	-0.35	-3.02	**0.004**	-0.88	0.43	-1.74	-0.02	-2.06	**0.045**
	Partner	0.29	0.37	-0.46	1.05	0.78	0.442	-0.26	0.38	-1.02	0.51	-0.67	0.505
Interaction effects:	NPI-Apathy	-1.04	0.30	-1.64	-0.44	-3.49	**0.001**	-0.48	0.33	-1.15	0.19	-1.46	0.153
	ZBI	-0.24	0.10	-0.45	-0.03	-2.27	**0.028**	-0.01	0.12	-0.24	0.22	-0.08	0.938
Model II: Relationship satisfaction		**DEPENDENT VARIABLE: DEPRESSION**											
	Actor	-0.15	0.06	-0.27	-0.04	-2.75	**0.009**	-0.12	0.05	-0.23	-0.02	-2.34	**0.024**
	Partner	0.01	0.06	-0.10	0.13	0.23	0.817	0.08	0.05	-0.03	0.18	1.45	0.155
Interaction effects:	NPI-Apathy	-0.16	0.13	-0.42	0.10	-1.23	0.223	0.17	0.12	-0.08	0.42	1.41	0.166
	ZBI	0.12	0.04	0.04	0.19	3.18	**0.003**	0.08	0.04	0.01	0.15	2.18	**0.035**
Model III: Anxiety		**DEPENDENT VARIABLE: RELATIONSHIP SATISFACTION**											
	Actor	-0.48	0.36	-1.21	0.26	-1.30	0.199	-0.88	0.25	-1.39	-0.38	-3.53	**0.001**
	Partner	0.02	0.28	-0.55	0.58	0.05	0.958	0.93	0.31	0.31	1.55	3.02	**0.004**
Interaction effects:	Age	0.33	0.17	0.00	0.67	2.00	0.052	0.26	0.14	-0.03	0.56	1.83	0.075
	NPI-Apathy	-0.95	0.32	-1.61	-0.30	-2.94	**0.005**	-0.24	0.28	-0.80	0.32	-0.87	0.391
	ZBI	-0.29	0.11	-0.51	-0.07	-2.61	**0.012**	-0.22	0.09	-0.42	-0.03	-2.39	**0.022**
Model IV: Relationship satisfaction		**DEPENDENT VARIABLE: ANXIETY**											
	Actor	-0.08	0.06	-0.21	0.04	-1.32	0.193	-0.22	0.08	-0.37	-0.06	-2.74	**0.009**
	Partner	0.14	0.06	0.01	0.26	2.19	**0.034**	0.03	0.08	-0.12	0.19	0.42	0.675
Interaction effects:	Age	0.02	0.07	-0.12	0.16	0.24	0.811	-0.05	0.09	-0.23	0.12	-0.61	0.546
	NPI-Apathy	-0.15	0.14	-0.43	0.13	-1.08	0.287	0.09	0.17	-0.27	0.44	0.50	0.618

	ZBI	0.19	0.04	0.12	0.27	4.92	**0.000**	0.04	0.05	-0.06	0.14	0.84	0.404
Model V: Quality of life						**DEPENDENT VARIABLE: RELATIONSHIP SATISFACTION**							
	Actor	0.19	0.08	0.04	0.34	2.57	**0.013**	0.15	0.06	0.02	0.28	2.37	**0.022**
	Partner	0.07	0.08	-0.09	0.22	0.88	0.386	-0.05	0.06	-0.17	0.07	-0.89	0.377
Interaction effects:	Age	0.47	0.19	0.10	0.85	2.54	**0.014**	0.39	0.16	0.07	0.70	2.48	**0.017**
	NPI-Apathy	-1.16	0.32	-1.80	-0.51	-3.59	**0.001**	-0.59	0.26	-1.11	-0.07	-2.29	**0.027**
Model VI: Relationship satisfaction						**DEPENDENT VARIABLE: QUALITY OF LIFE**							
	Actor	0.56	0.27	0.03	1.10	2.13	**0.039**	0.75	0.32	0.12	1.39	2.38	**0.021**
	Partner	-0.41	0.32	-1.06	0.24	-1.27	0.210	-0.09	0.24	-0.58	0.41	-0.36	0.724
Interaction effects:	Age	-0.24	0.38	-1.00	0.52	-0.64	0.527	0.51	0.37	-0.24	1.25	1.36	0.179
	NPI-Apathy	0.07	0.72	-1.39	1.53	0.10	0.925	0.42	0.68	-0.94	1.78	0.62	0.537

Abbreviations: B – unstandardised coefficient estimates; CI – confidence interval; NPI – Neuropsychiatric Inventory; SE – standard error; ZBI – Zarit Burden Interview.

Figure 8.2 Feedback cycle between the people with PRD's anxiety and relationship satisfaction of life partners and people with PRD.

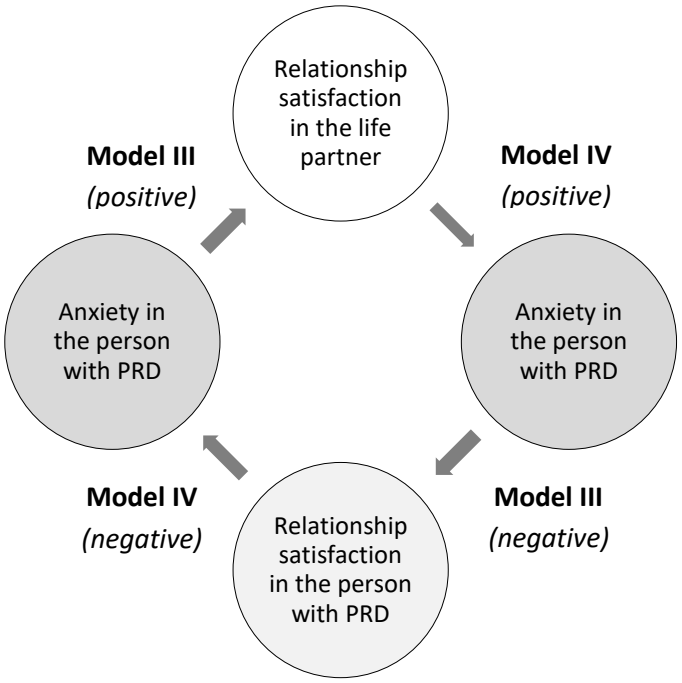
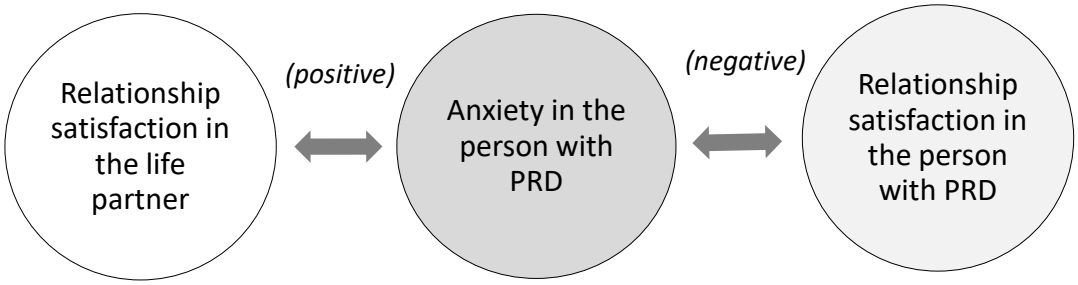


Figure 8.3 Bidirectional relationship between the people with PRD's anxiety and relationship satisfaction of life partners and people with PRD.



8.5 Discussion

8.5.1 General discussion

To my knowledge, this is the first study to jointly explore the associations of health-related outcomes and relationship satisfaction among life partners and people with Parkinson's-related dementia. The majority of couples in this study comprised males with PRD and female life partners, which is representative of couples where one of the partners has a PD diagnosis (Cifu et al., 2006; Hand et al., 2018; Martinez-Martin et al., 2015). Life partners were significantly less satisfied with the relationship than people with PRD, with around half of life partners reporting relationship dissatisfaction compared to one third of people with PRD. This is a novel finding emerging from this study, as mutuality scores did not differ between couples where one partner had PD (Karlstedt et al., 2017).

Lower relationship satisfaction in both partners was significantly associated with health-related outcomes (i.e. higher depression and anxiety and lower quality of life) and socio-demographic aspects (i.e. shorter relationship duration and younger age) of both partners as well as with higher life partners' burden, which is consistent with earlier studies in dementia and PD (Fauth et al., 2012; Morris et al., 1988; Steadman et al., 2007; Tanji et al., 2008).

The APIM, when adjusted for covariates and corrected for multiple comparisons, revealed that there were significant actor effects between depression and relationship satisfaction among life partners and between anxiety and relationship satisfaction among people with PRD. In addition, a partner effect emerged between anxiety scores of people with PRD and relationship satisfaction of life partners, which is somewhat unexpected. However, it may be explained by the fact that life partners may become more concerned when their partner is exhibiting neuropsychiatric symptoms such as anxiety and may therefore seek more proximity and closeness with the care recipient to support them. When the APIM was not

corrected for multiple comparisons, an additional partner effect emerged whereby relationship satisfaction of life partners predicted anxiety among people with PRD. This could be explained by the fact that when life partners are more satisfied with the relationship, people with PRD may become anxious because they may feel they might be unable to fulfil the expectations of life partners in terms of intimacy, closeness and mutuality due to their health and PD-related symptoms as they used to be able to before the PD diagnosis. These findings, however, should be interpreted with caution due to the small sample size, exploratory nature of the study and low rates of anxiety and depression among participants and further studies are required to establish the links between health-related outcomes and relationship satisfaction in this population.

The actor effects of depression among life partners and people with PRD on relationship satisfaction found in this study build on earlier research conducted with healthy married couples (Whisman, Uebelacker, & Weinstock, 2004). In another study with married couples outside of PD and PRD, higher anxiety scores in wives were associated with positive relationship quality (but not negative relationship quality) among husbands, which the authors explained by how marital quality was measured: positive relationship quality focused on whether support behaviour existed and negative relationship quality explored whether there were conflicts in the relationship (Zaider, Heimerg, & Iida, 2010). Thus, the authors concluded that when women had high scores of anxiety, their husbands received less support from their spouses but husbands did not necessarily experience disharmony or discord in the relationship (Zaider et al., 2010). The association in the current study between higher life partners' relationship satisfaction and higher person with PRD's anxiety highlights that the association between the outcomes of both members of the dyad in PRD is complex and needs further research.

It is noteworthy that a small proportion of people with PRD in this study had clinically significant depression and anxiety. In an earlier study, 17% of people with PD had depression (Reijnders, Ehrt, Weber, Aarsland, & Leetntjens, 2008) which was higher compared to the current study (i.e. 10%) but anxiety levels have been

reported between 3.5% to 57% of people with PD (Broen, Narayen, Kuijf, Dissanayaka, & Leentjens, 2016; Dissanayaka et al., 2010, 2014) which was comparable to the current study as 20% of people with PRD reported clinically significant anxiety. An earlier study with 87 people with PD found that nearly 30% experienced fluctuations in anxiety (Richard et al., 2004) which could explain the relatively low levels of anxiety reported by people with PRD in the current study. Once people with PD have developed dementia, the levels of anxiety and depression increase (Martinez-Martin et al., 2015), which is associated with higher life partners' burden, similarly to earlier findings (Oh et al., 2015). Among care partners of people with PD, female care partners tend to experience higher levels of anxiety and depression than male care partners (Gultekin, Ekinci, Erturk, & Mirza, 2017), which is an increasing trend once dementia has emerged in PD, although the scores were not statistically different between PD-normal cognition, PD-MCI and PDD (Lawson et al., 2017). Nevertheless, this highlights that cognitive impairment in PD may be a significant stressor for life partners and can directly impact their well-being and health outcomes.

The relatively low proportion of participants in the study experiencing depression and anxiety could be explained by: (1) the inclusion of people with PD-MCI, PDD and DLB collectively, where there might be significant variability in the sample as people with PD-MCI have lower levels of anxiety and depression than people with PD-related dementia (Martinez-Martin et al., 2015), (2) including participants who were recruited for the pilot study of a psychosocial intervention (i.e. INVEST study) who may have been more motivated to participate in a research study and thus who may have had lower levels of depression, anxiety and apathy compared to the wider population of people with PRD who may not have been interested or motivated to participate in the study, (3) people with PRD may have other prominent neuropsychiatric symptoms, such as apathy, whereas people with PD without cognitive impairment may have more depression and anxiety (Martinez-Martin et al., 2015), and (4) collecting data at one time-point only which provides a snapshot of the mental health levels of participants, which may fluctuate, increase or decrease or improve spontaneously depending on the circumstances, symptom

presentation of PD and/or whether the person with clinically significant anxiety or depression is receiving or has received pharmacological or non-pharmacological treatment. This suggests that a study with a larger cohort of participants with each clinical syndrome within PRD may need to be conducted to address these aspects.

Exploring the associations between health-related outcomes and relationship satisfaction in both partners in PRD is important because being married (Hakansson et al., 2009; Xu et al., 2016) and having higher relationship closeness can both protect against cognitive impairment in later life or slow down the progression of cognitive impairment in the person with dementia (Norton et al., 2009). Furthermore, it is noteworthy that relationship satisfaction remains relatively high among older adults (Braun, Rohr, Wagner, & Kunzmann, 2018) and is related to health and well-being (Margelisch, Schneewind, Violette, & Perrig-Chiello, 2017), which the findings of the current study supported. In contrast, relationship dissatisfaction can contribute to poor well-being and mental health which highlight that focusing on supporting dyadic relationships is crucial in PRD. The results from this study suggest that relationships are more complex and multifaceted due to one of the partners living with a neurodegenerative condition such as PD-MCI, PDD or DLB and it is suggested that future research be conducted to study the associations in more depth in a larger cohort of couples.

8.5.2 Methodological strengths

This is the first study to explore the associations between outcomes of people with PRD and life partners jointly and demonstrated that there exist actor and partner effects between specific outcomes, which have not been described before in the context of intimate relationships in PRD. There is only one other study of APIM in PD (i.e. Mavandadi et al., 2014) and the current study is an important and novel addition into expanding the field of a complex type of dementia such as PDD and DLB. Including both members of the couple in one model is considered a strength because it makes it possible to explore the mechanisms and associations between health-related outcomes and relationship satisfaction, and to explore how people

with PRD and life partners can mutually impact and influence each other. Furthermore, the majority of studies in PD and PRD have not taken into account the outcomes of both partners in one model and the current study demonstrated that several partners' outcomes are interconnected, which is a new finding emerging from this study. Another key strength is the inclusion of people with three clinical syndromes in PD-related cognitive impairment, which differs from many other studies who have only included people with PDD and DLB or people with PD-MCI and PDD.

8.5.3 Limitations

Despite the numerous strengths, there were some limitations. This study applied several statistical analyses with a relatively small sample size of 57 couples; thus, the statistical power was relatively low. There could have also been an inflated rate of type I error whereby the findings could have falsely indicated a significant result which in reality may not exist and it is essential to bear this in mind when interpreting the findings. One way this was addressed in the study was by applying a Bonferroni post hoc test and presenting the findings with a more stringent level of significance (i.e. $p = 0.0125$). However, a less stringent p-value cut-off was selected due to the exploratory nature of the study and the imperative not to miss a promising signal which would drive future research questions. Thus, it was vital to examine all possible associations among the variables so as to give an insight into the variables of interest in this population. To increase power, which is one way of reducing the risk of a Type I error, the study should be replicated with a larger sample size (Forstmeier, Wagenmakers, & Parker, 2017). Furthermore, despite the small sample size in the study, it was higher than in similar earlier studies with older people (Mavandadi et al., 2014; Regan et al., 2014; Walker, Isherwood, Burton, Kitwe-Magambo, & Luszcz, 2013).

The gender imbalance in the sample (i.e. more men with PRD than women with PRD, and more female life partners than male life partners) limits the generalisability of the findings to a wider population of people with PRD and life

partners. Similarly, only a small proportion of people with PRD and life partners in this study had clinically significant anxiety and depression and this might have precluded drawing accurate conclusions about the links between the studied variables and relationship (dis)satisfaction. The sample may also not have been representative of a typical sample of people with PRD among whom the frequency of depression and anxiety may be higher than that reported in the current study (Dissanayaka et al., 2010, 2014; Reijnders et al., 2008). In order to obtain more accurate estimates about the study variables, it would be important to include more people with clinically significantly anxiety or depression in the study.

Another limitation is the cross-sectional nature of the study, which limits the possibility of drawing conclusions regarding causality of the studied variables. Employing a longitudinal design using APIM would help in understanding more about the cause and effect of the associations between the variables. All participants in this study completed self-reported measures which may not reflect objective health outcomes, particularly among people with PRD who may under- or over-report neuropsychiatric symptoms and who may have more fluctuations in terms of neuropsychiatric symptoms. To attain accurate scores of health outcomes among participants, self-reported and observer- or clinician-rated outcomes could be used concurrently, which would potentially provide a more comprehensive overview of symptoms among people with PRD. Finally, only a small number of measures was included in the study which prevented a further exploration of associations between relationship satisfaction, relationship quality, mutuality, dyadic support, coping and other health-related variables such as apathy and stress in both partners.

8.5.4 Future directions

A future study should recruit a larger sample of people with PD-MCI, PDD and DLB and life partners so that the models would be more robust. This would also allow the conduct of structural equation modelling to observe the potential moderating and/or mediating effects between the studied outcomes. Furthermore, an

important avenue of research would be to examine the impact of different diagnostic subtypes or stages (mild, moderate, advanced) of dementia on key dyadic outcomes, including measures of apathy, stress, coping mechanisms, dyadic support, relationship quality, and mutuality, as well as employing a longitudinal study design to observe the causal effects in more detail. As intimate relationships are interpersonal, complex and multifaceted, future studies could potentially combine quantitative and qualitative methods to depict a more accurate picture of relationships in PRD.

Focusing on the mental health and well-being, including prevention and treatment of anxiety and depression, in people with dementia and their care partners is one of the priority areas for research in the 'quality of life' domain identified by the UK's Alzheimer's Society "Dementia research roadmap" (Alzheimer's Society, 2018). Additionally, the Alzheimer's Society aims to develop viable ways of supporting all those who are affected by dementia, recognise the heterogeneity of care partners within families and social circles, and acknowledge the dynamics and relationships between the care recipient, care partner and the family. Thus, it is important and timely to focus on dyadic studies and interventions.

8.6 Conclusion

The current study is the first to explore the associations between relationship satisfaction and health-related outcomes of people with PRD and life partners jointly and is an important step forward in understanding the dyadic influences of relationship (dis)satisfaction among couples. This study evidenced that health-related outcomes (i.e. anxiety, depression, quality of life) and relationship satisfaction are associated in both partners; however, it is not possible to ascertain the presence of bidirectional relationship between the studied variables. Importantly, life partners are less satisfied with the relationship than people with PRD and there may be a role for psychosocial interventions that support and improve mental health, quality of life and well-being of each individual that

ultimately contribute to maintenance and sustenance of intimate dyadic relationships in PRD.

CHAPTER 9: Does Cognitive Stimulation Therapy in Parkinson's-related dementia improve life partners' relationship satisfaction, burden and well-being? An exploratory secondary analysis of a pilot randomised controlled trial (Study 5)

The study described in this chapter is part of the INVEST RCT paper and the following manuscript has been submitted:

Leroi, I., **Vatter, S.,** Carter, L.-A., Smith, S. J., Orgeta, V., … McCormick, I. (under review). Parkinson's-adapted Cognitive Stimulation Therapy: A pilot randomised controlled clinical trial. *Therapeutic Advances in Neurological Disorders*

This chapter describes a secondary analysis of the INVEST study, a single-blind two-arm pilot feasibility randomised controlled trial of Cognitive Stimulation Therapy in Parkinson's-related Dementia (CST-PD). The chapter follows the extensions of the Consolidated Standards of Reporting Trials (CONSORT) statements to: (1) randomised pilot and feasibility trials (Eldridge et al., 2016b), and (2) assessing non-pharmacological treatments (Boutron et al., 2017) to maximise transparency and quality of reporting the method and results of the study.

9.1 Introduction

PD-MCI, PDD and DLB are complex and progressive conditions within Lewy body spectrum disorders LBSD requiring regular help and support from a care partner. Commonly the role of a care partner is fulfilled by a life partner or a spouse. However, providing care to a care recipient can result in increased anxiety and depression (Martinez-Martin et al., 2008; Schrag et al., 2006), burden and stress (Carter et al., 2008; Leiknes et al., 2015; Martinez-Martin et al., 2008; Mosley et al., 2017), reduced life satisfaction (Aarsland et al., 1999a) and lower quality of life (Lawson et al., 2017; Leroi et al., 2012a; Martinez-Martin et al., 2008) for life partners of people with Parkinson's-related dementia (PRD). Life partners can also

be older adults themselves and may have physical and mental health issues raising challenges in managing their own health and their partners' health simultaneously, on a daily basis. This highlights the need to focus on life partners when undertaking interventions to improve well-being and to support coping.

Non-pharmacological interventions have gained more interest among researchers in the last decades due to their benefit, efficiency, individualised approach, minimal harm, bypassing any drug-induced side effects and cost-effectiveness (Goldman et al., 2018). Frequently, non-pharmacological interventions are targeted at people with a specific condition but the evidence-base for non-drug-based options for care partners of people with a complex disorder is growing. Zarit (2018) highlighted the gap in knowledge regarding care partner interventions to improve long-term outcomes and argues for more research. To date, several psychological interventions have been trialled with care partners of people with dementia. The most popular interventions have been psychosocial therapies, psychoeducational approaches, various types of therapies (i.e. occupational, talking, cognitive behavioural therapy), technological, support group and multicomponent interventions as summarised in a meta-review (Gilhooly et al., 2016). These interventions are delivered individually, in a group, online or as a dyad with the person who has dementia. From these non-pharmacological approaches, psychosocial and psychoeducational therapies were considered to be of highest benefit to care partners (Gilhooly et al., 2016), which when tailored and delivered face-to-face with skills training, can increase the intervention's effectiveness (Weinbrecht et al., 2016). While this evidence is encouraging, the applicability of such interventions to less common forms of dementia remains unclear. In particular, the evidence to support non-pharmacological interventions for life partners of people with Parkinson's disease dementia (PDD; Hindle et al., 2013) and dementia with Lewy bodies (DLB; Connors et al., 2018) is almost non-existent. Adapting existing interventions specifically for this group, and learning from previous experience may help in finding an effective intervention for this population.

Psychosocial interventions are most often designed to target burden, stress, depression, quality of life, mental well-being, social support and relationship satisfaction among care partners (Abrahams et al., 2018; Gilhooly et al., 2016; Hindle et al., 2018; Hopwood et al., 2018; Kwon et al., 2017; Laver, Milte, Dyer, & Crotty, 2017; Orrell et al., 2017). However, relatively few studies have focused on exploring specific aspects of the care relationship, such as interaction, positive feelings related to care provision and relationship satisfaction and a deeper understanding of these aspects is required. In a recent systematic review, the authors concluded that for people with dementia, quality of life can be improved by supporting relationships with loved ones and encouraging social participation (Martyr et al., 2018), which can also apply to life partners and can be addressed by providing support specifically directed at the care provider-recipient dyad.

Dyadic interventions for people with dementia have most often targeted cognition and are typically divided into cognitive rehabilitation, cognitive training and cognitive stimulation (Bahar-Fuchs, 2013). As there is an urgent need to focus on rigorous, evidence-based, controlled trials for people with PRD and their care partners, pilot RCTs of each therapy are being conducted to determine their effectiveness in this population. Hindle and colleagues (2018) have completed a goal-oriented cognitive rehabilitation trial with people with PRD and their care partners and found that care partners' quality of life and health status improved after receiving the intervention. In a cognitive training intervention for people with Parkinson's disease and mild cognitive impairment (PD-MCI), care partners received an education programme (Reuter, Mehnert, Sammer, Oechsner & Engelhardt, 2012) which brought them benefits in terms of feeling more relaxed and being capable of coping with various situations. However, the effectiveness of cognitive training in people with PDD and DLB and their care partners is yet to be determined (Orgeta et al., 2015b; Orgeta et al., under review). A trial of individual Cognitive Stimulation Therapy (iCST; Orrell et al., 2017) amongst people with dementia and their care partners showed that care partners' quality of life improved as well as the perceptions of relationship quality held by the person with dementia. CST has been specifically adapted to meet the needs of people with PRD (CST-PD, the INVEST

trial; McCormick et al., 2017a) and the trial was recently undertaken with dyads of people with PRD and their care partners (Leroi et al., under review; McCormick et al., 2017b; McCormick et al., in press). The primary aim of the INVEST study was to evaluate feasibility of recruitment, acceptability and tolerability of the intervention, as well as to explore the efficacy of CST-PD in improving cognitive impairment and quality of life in people with PRD. Collectively, an understanding of these operational aspects and the exploratory evaluation will provide the necessary information for a subsequent full-scale RCT trial (McCormick et al., 2017b). The secondary aim in the INVEST study was to evaluate whether the intervention improved outcomes in people with PRD, whereas the focus of the current study was to conduct a secondary analysis of outcomes in life partners. Specifically, the current study aimed to determine whether CST-PD had any effect on relationship satisfaction, burden and well-being in life partners.

9.2 Aim

The main objective of the current study was to conduct a secondary analysis of the INVEST study to investigate whether life partners' satisfaction with the relationship with the person with PRD was higher in the experimental group (Cognitive Stimulation Therapy for Parkinson's-related Dementia, CST-PD) compared to the control group (treatment as usual, TAU). Additionally, secondary outcome measures, such as life partners' burden, mental health and quality of life were explored in each arm.

9.3 Methods

Chapter 4 provides a detailed overview of the design, eligibility criteria, recruitment, procedure, data collection, randomisation, blinding, outcome measures, CST-PD intervention and ethics of the INVEST study. The current chapter briefly summarises the methods of this study.

9.3.1 Research design

The INVEST study was a phase II (exploratory) two-arm (CST-PD versus TAU) single-blind pilot RCT, which followed the guidelines of the Medical Research Council (MRC) framework for developing and evaluating complex interventions (Craig et al., 2008). Recruited dyads had a 1:1 chance to be randomly allocated to twelve weeks of either CST-PD (experimental arm) or TAU (control arm). Informed written consent was obtained from all participants prior to participation and all participants had the capacity to consent to participation.

9.3.2 Participant sample and recruitment

Participant-dyads were recruited to the INVEST trial from seven sites between 12/04/2016 and 31/07/2017. The final follow-up assessments were completed on 24/11/2017. Specific details of the recruitment sites and strategy are provided in Chapter 4, Sections 4.2.2.3 and 4.2.2.4, respectively.

9.3.2.1 Power analysis

Taking into consideration that this is a secondary analysis, the sample size in this study comprised of all eligible participants from the INVEST study (n = 57 life partners). However, to estimate the required sample size, a power analysis was conducted in GPower (version 3.1.9.2) which showed that for an analysis of covariance (ANCOVA) with a medium effect size of 0.25 (Cohen, 1988), alpha = 0.05 and power = 0.80, the projected sample size is 128 participants.

9.3.3 Procedure

Life partners completed sociodemographic and clinical assessments during screening and baseline visits (described in Chapter 4, Sections 4.2.3.4, 4.2.3.5 and 4.2.4.1). On completion of the baseline assessments, participants were randomised either to 12-week CST-PD or TAU. Follow-up assessments were completed after

participation in the trial had ended. The researchers undertaking baseline and follow-up visits remained blinded throughout the data collection period.

9.3.4 Measures

Life partners completed a battery of assessments (described in detail in Chapter 4, section 4.2.4.1) of which five scales were included for this chapter:

- The Relationship Satisfaction Scale (RSS; Burns, 1983),
- Dyadic Relationship Scale (DRS) positive interaction sub-scale (Sebern & Whitlatch, 2007),
- The Zarit Burden Interview (ZBI; Zarit et al., 1980),
- The Short Form 12 Health Survey (SF-12; Ware et al., 1996), and
- The EuroQoL-5D-3L (EQ-5D; The EuroQol group, 1990).

9.3.5 Analyses

Baseline characteristics of the sample were described using means, standard deviations and ranges for continuous data, and counts and percentages for categorical data. Comparisons between baseline characteristics of CST-PD and TAU groups were not undertaken as the groups were independent from each other and any differences between the groups that may have arisen are random and should not be tested (CONSORT 2010 guidelines, Moher et al., 2010). Baseline and follow-up variables were evaluated for normal distribution of the data with skewness and kurtosis z-scores and Shapiro-Wilk's normality test.

An intention-to-treat (ITT) analysis was applied and all participant data were included according to the randomisation (Fisher et al., 1990). An ITT, referred to as "once randomised, always analysed" (Schulz & Grimes, 2002), is a preferred data analysis method as it preserves sample size, removes bias and assures participants' data are maintained and analysed as randomised (Gupta, 2011; Hollis & Campbell, 1999; Polit & Gillespie, 2010; Ranganathan, Pramesh & Aggarwal, 2016; Schulz,

Altman, Moher & CONSORT group, 2010; Schulz & Grimes, 2002; White, Carpenter & Horton, 2012). An ITT disregards withdrawal from the trial, protocol deviations, eligibility violations identified after randomisation, non-compliance and non-adherence to the intervention or trial (Fisher et al., 1990; Gupta, 2011). A traditional, non-modified ITT requires 0% attrition or the replacement of any outcome values that are missing (Polit & Gillespie, 2010).

In line with the ITT principle, the missing data values were imputed. In order to decide how to address the missing values in the dataset, Little's Missing Completely at Random (MCAR; Little, 1988) test was carried out in SPSS. The Little's MCAR chi-square test result [(1023, N = 57) = 368.22, p = 1.000] revealed that the data were missing at random, meeting the assumption for undertaking an imputation method (i.e. multiple imputation) to populate missing values. In this study some missing information occurred in 7% of total values, in 65.6% of all the variables and in 30 participants from a sample of 57 (of whom 10 had withdrawn or were lost to follow-up). On a variable level, missing data occurred between 1.8% and 24.6% of cases (between 1 and 14 missing values per variable, respectively). There was a monotone pattern of missingness as assessed in the visual inspection of the missing value patterns matrix (Appendix K).

All missing data were imputed with multiple imputation, which assumes the data are missing at random (Nguyen, Carlin & Lee, 2017). In the current study, two dyads did not wish to continue participating in the intervention arm due to finding it hard to engage the person with PRD in the therapy indicating a non-random missingness; however, as it only occurred in two dyads, their data were included under the 'missing at random' assumption. In multiple imputation, "*multiple sets of plausible values for missing data were created from their model-based predictive distribution, and estimates and standard errors were obtained with the use of multiple-imputation combining rules"* (Little et al., 2012, p. 1358). Specifically, the missing data are predicted using regression analysis according to the values in the dataset and replaced with those values that were predicted creating a complete dataset (Kang, 2013; Sinharay, Stern & Russell, 2001). Multiple imputation does not aim to

generate additional data but to compute and replace the missing values in order to perform data analysis on the full data set (Barnes, Lindborg & Seaman Jr, 2006).

Multiple imputation has been found to perform well with non-normal data and small sample sizes (~ 50 participants) and produce correct variability in comparison to single imputation (Graham, 2009; Graham & Schafer, 1999; Kang, 2013). The process of imputing values with multiple imputation involves three steps: *"(1) the missing data are filled in m times to generate m complete data sets, (2) the m complete data sets are analysed by using standard procedures, and (3) the results from the m complete data sets are combined for the inference*" (Yuan, 2010, p. 1). The missing items were imputed on a scale-level, rather than item-level, as (1) there is generally little difference between scale- and item-level imputation with regards to the bias they introduce to scale-level estimates (although item-level imputation produces better power), and (2) imputation at item-level cannot be undertaken due to small sample size (i.e. the number of cases should be similar to or higher than the number of variables) (Gottschall, West & Enders, 2012). The model of multiple imputation included the sociodemographic variables (gender, age, ethnicity and marital status of both partners), participant and life partner related factors (e.g. disease duration, care provision duration) and outcome scores at baseline and follow-up. Following the general rule of thumb to apply between 3 and 10 imputations (Rubin, 1987), seven imputations were applied which was directly related to the percentage of missing information in the current dataset (i.e. 7%) (Bodner, 2008; Graham, Olchowski & Gilreath, 2007; Von Hippel, 2009).

For the purposes of evaluating the outcome measures, a one-way ANCOVA was undertaken, whereby the follow-up outcome measure was the dependent variable, group allocation was a fixed factor (independent variable) and the analyses was adjusted for baseline scores (i.e. the covariates). All statistical analyses were conducted in the Statistical Package for the Social Sciences (SPSS; version 23). The level of statistical significance was set at $p < 0.05$.

Prior to undertaking ANCOVA, the assumption tests were evaluated for each outcome measure via statistical tests and visual inspection of the graphs (described in detail in Appendix K).

9.4 Results

9.4.1 Participant recruitment (INVEST)

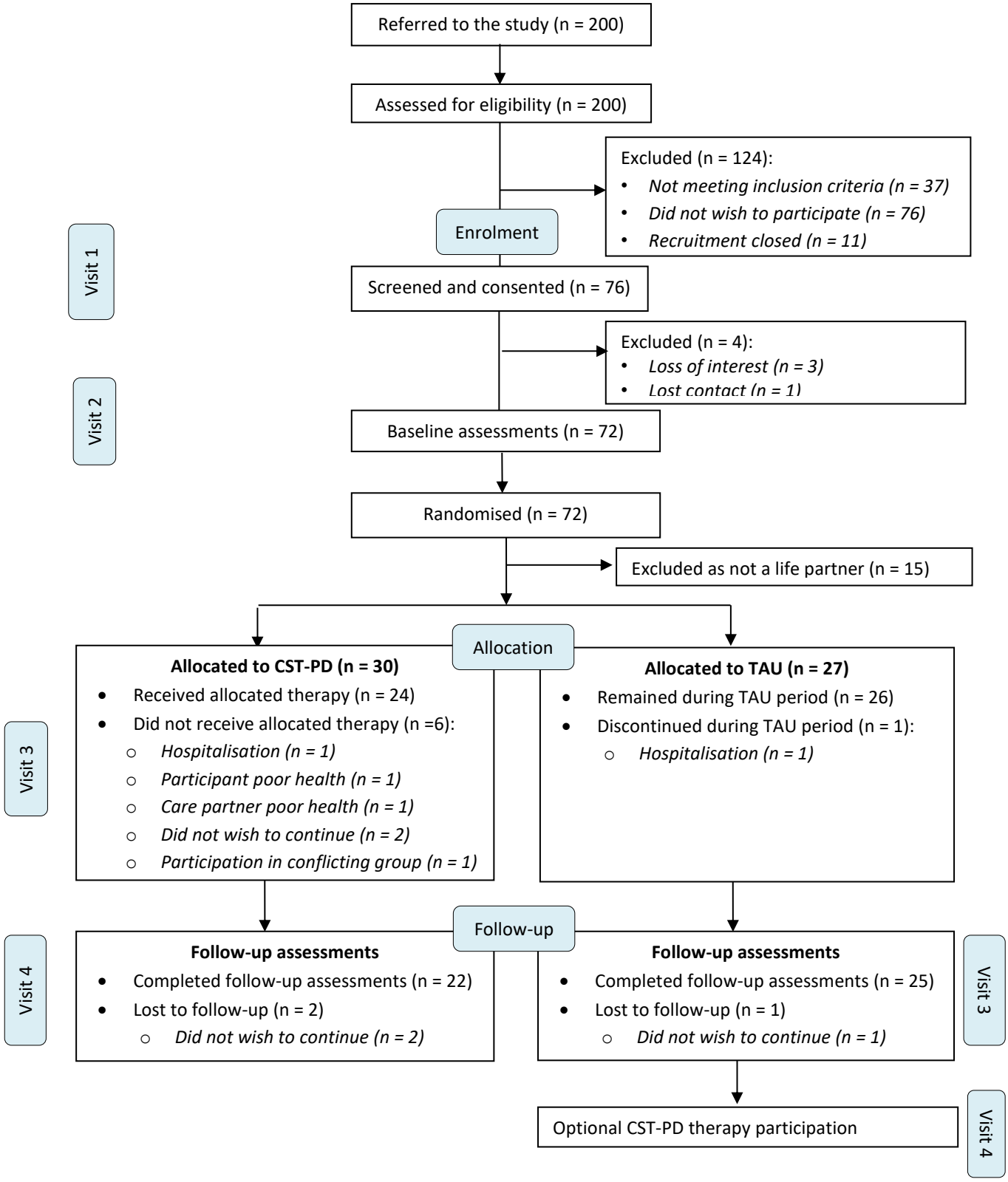
Across the seven recruiting sites, 200 dyads were referred of whom 37 were ineligible, 11 were referred after recruitment to the study had closed and 76 were not interested in participating. Recruitment took place between April 2016 and July 2017 with final follow-up assessments completed in November 2017. A total of 76 dyads provided informed consent and were recruited to the INVEST study (summary provided in Table 9.1). Four dyads were lost after recruitment due to lack of interest in participating (n = 3) and loss of contact with the dyad (n = 1); therefore, 72 dyads were randomised, received intended treatment (CST-PD or TAU) and were assessed for each objective. The recruitment rate (number of participants recruited per site per month) in the INVEST study was 38% and attrition rate 26% (number of participants who discontinued participation in the study). PAT (59.1%) and NELFT (57.1%) had the highest conversion rates from dyad referral to recruitment. The reasons for ineligibility included no diagnosis of PD (n = 8) or cognitive impairment (n = 6), distance (n = 3), admitted to care home (n = 2), impaired health (n = 15) and study partner or consultee not identified (n = 3). The reasons for non-participation included worsening health and dementia in the participant (n = 3), care partner burden (n = 6), failure to contact the dyad (n = 11), lack of time to commit (n = 3), unwillingness to be randomised (n = 1) and lack of interest in participating (n = 52).

Table 9.1 Participant progression in the INVEST trial.

Sites	Referred	Ineligible	Not interested or other reason	Recruitment closed	Recruited	Randomised & received CST-PD or TAU
GMMH	58	18	21	3	16	15
SRFT	23	3	9	1	10	8
PAT	22	2	7	0	13	13
UHSM	26	4	14	1	7	6
Derbyshire	29	3	11	3	12	12
NELFT	21	3	6	0	12	12
NWBH	21	4	8	3	6	6
Total	200	37	76	11	76	72

Abbreviations: CST-PD – Cognitive Stimulation Therapy in Parkinson's-related dementia; Derbyshire – Derbyshire Healthcare Trust; GMMH - Greater Manchester Mental Health NHS Foundation Trust; NELFT – North East London Foundation Trust; NWBH - North West Boroughs Healthcare Trust; PAT - Pennine Acute Trust; SRFT - Salford Royal Foundation Trust; TAU – treatment as usual; UHSM – University Hospital of South Manchester.

Figure 9.1 CONSORT flow diagram for dyads.



9.4.2 Participant flow (current study)

For the purposes of this study only the data for eligible life partners was included (n = 57). The primary analysis was intention-to-treat which included all participants according to randomisation. The flow of dyads through the trial is seen in Figure 9.1. Ten dyads withdrew over the course of the study (eight in the therapy arm and two in the control arm) and did not complete follow-up assessments. The main reasons for withdrawal were due to hospitalisation or poor health (n = 4) and not wishing to continue (n = 5). The blinded researchers correctly guessed the allocation for 41.9% of dyads in the therapy arm and 61.3% dyads in the control arm.

9.4.3 Participant baseline demographic and clinical characteristics

Characteristics of life partners and people with PRD for the overall sample and across randomisation arms are provided in Table 9.2. Overall, the mean age of life partners was 69.5 years (SD = 6.94). Life partners were predominantly female (91.2%), white British (91.2%) and married (93%). All couples lived together and the average duration of the relationship was 45.4 years (SD = 12.80). In terms of education, over half of the participants (52.6%) had left school between the ages of 14 and 18, a quarter of life partners (24.6%) had completed further education and 13 participants (22.8%) had an undergraduate or postgraduate degree. On average, life partners had provided care for 5.3 years (SD = 4.64) and were currently providing care for 98.8 hours a week (SD = 66.57).

People with PRD were mostly male (91.2%) and white British (93%), with a mean age of 74 years (SD = 6.64). Around half of the people with PRD were diagnosed with PDD (43.8%), followed by PD-MCI (31.5%) and DLB (24.6%). People with PRD had Parkinson's disease and cognitive impairment on average for 8.4 years (SD = 7.10) and 3.9 years (SD = 3.53), respectively. The mean MoCA score for people with PRD was 18.40 (SD = 5.39). Similarly to life partners, half (50.9%) of the people with PRD had completed secondary education, 26.3% further education and 22.8% higher education (i.e. a university degree).

Table 9.2 Participant characteristics at baseline (n = 57 life partners).

	Life partners			People with PRD		
Categorical variables N (%)	**Overall (n = 57)**	**CST-PD (n = 30)**	**TAU (n = 27)**	**Overall (n = 57)**	**CST-PD (n = 30)**	**TAU (n = 27)**
Gender, female	52 (91.2)	26 (45.6)	26 (45.6)	5 (8.8)	4 (7.1)	1 (1.8)
Ethnicity, white British	52 (91.2)	27 (47.4)	25 (43.8)	53 (93.0)	29 (50.9)	24 (42.1)
Relationship status, married	53 (93.0)	27 (47.4)	26 (45.6)			
Living status, with spouse/ partner	57 (100)	N/A	N/A			
Education						
Left school aged 14-16	21 (36.8)	9 (15.8)	12 (21.0)	26 (45.6)	13 (22.8)	13 (22.8)
Left school aged 17-18	9 (15.8)	5 (8.8)	4 (7.0)	3 (5.3)	0	3 (5.3)
Further education	14 (24.6)	7 (12.3)	7(12.3)	15 (26.3)	9 (15.8)	6 (10.5)
Higher education	9 (15.8)	6 (10.5)	3 (5.3)	6 (10.5)	3 (5.3)	3 (5.3)
Postgraduate	4 (7.1)	3 (5.3)	1 (1.8)	7 (12.3)	5 (8.8)	2 (3.5)
Diagnosis						
PD-MCI				18 (31.5)	10 (16.7)	8 (14.8)
PDD				25 (43.8)	12 (21)	13 (22.8)
DLB				14 (24.6)	8 (14)	6 (10.5)
H&Y						
Stage 1.0				5 (9.1)	3 (5.5)	2 (3.6)
Stage 1.5				4 (7.2)	2 (3.6)	2 (3.6)
Stage 2.0				17 (30.9)	8 (14.5)	9 (16.4)
Stage 2.5				11 (20.0)	7 (12.7)	4 (7.3)
Stage 3.0				6 (10.9)	4 (7.3)	2 (3.6)
Stage 4.0				10 (18.2)	4 (7.3)	6 (10.9)
Stage 5.0				2 (3.6)	1 (1.8)	1 (1.8)
Continuous variables Mean (SD) [range]						
Age, years	69.5 (6.94) [48-85]	67.8 (6.49) [48-78]	71.4 (7.02) [58-85]	74.0 (6.64) [55-90]	73.0 (6.44) [55-83]	75.0 (6.81) [61-90]
Relationship duration, years	45.4 (12.80) [10-61]	42.5 (13.76) [10-60]	48.6 (11.00) [10-61]			
Age left full-time education	17.2 (2.88) [14-31]	17.5 (3.42) [14-31]	16.9 (2.14) [15-24]	18.2 (5.31) [14-40]	18.5 (5.83) [14-40]	17.9 (4.75) [14-30]
MoCA score (max 30)	27.9 (2.00) [22-30]	27.8 (2.02) [23-30]	27.9 (2.00) [22-30]	18.40 (5.39) [7-30]	18.6 (5.65) [8-30]	18.1 (5.20) [7-24]
Duration of PD, years				8.4 (7.10) [1-37]	8.4 (6.88) [1-28]	8.5 (7.56) [1-37]
Duration of MCI / dementia, years				3.9 (3.53) [0.2-16]	4.3 (3.84) [0.8-14]	3.4 (3.16) [0.2-16]
UPDRS-III (max 100)				30.8 (12.66) [10-58]	29.6 (12.86) [10-58]	32.0 (12.5) [10-51]
SE-ADL (max 100)				52.9 (24.62) [10-90]	51.4 (25.60) [10-90]	54.4 (23.91) [20-90]
Care provision duration, years	5.3 (4.64) [0-20]	5.7 (5.18) [0-20]	4.8 (3.98) [0-15]			
Care provision hours/week	98.8 (66.57) [0-168]	97.9 (65.78) [0-168]	99.9 (68.65) [0-168]			
RSS (max 42)	27.4 (11.67) [0-42]	24.8 (12.82) [0-42]	30.2 (9.69) [10-42]			

Abbreviations: CST-PD – Cognitive Stimulation Therapy in Parkinson's-related Dementia; DLB – Dementia with Lewy bodies; MoCA – Montreal Cognitive Assessment; PD – Parkinson's disease; PDD – Parkinson's disease dementia; PD-MCI – Parkinson's disease and mild cognitive impairment; PRD – Parkinson's-related dementia; RSS – Relationship Satisfaction Scale; SE-ADL – Schwab & England Activities of Daily Living scale; TAU – treatment as usual; UPDRS – Unified Parkinson's Disease Rating Scale.

9.4.4 Analysis of covariance (ANCOVA)

Prior to conducting an analysis of covariance with each follow-up outcome as the dependent variable, tests of assumptions were undertaken (Appendix K).

The unadjusted means for each outcome for the two groups (CST-PD and TAU) at baseline and follow-up time-points are provided in Table 9.3.

After controlling for baseline relationship satisfaction score (RSS), there was no statistically significant difference between the CST-PD and TAU groups for relationship satisfaction at follow-up with the original data [F(1,43) = 3.375, p = 0.073, mean difference = 3.28, 95% CI = -0.32, 0.69, p = 0.073) or with the imputed data (mean pooled difference = 2.35, p-value varied between 0.082 and 0.308) (Table 9.4). However, the estimated marginal means of RSS indicated that relationship satisfaction was higher in the CST-PD arm (M = 30.07) in comparison to the TAU arm (M = 26.78) following the intervention, pointing to a positive trend of improvement on this measure. Furthermore, RSS significantly improved (mean difference = 3.46, 95% CI = -0.17, -6.75, p = 0.040) when the sample size was higher including all 76 care partners, whether or not they were life partners (Leroi et al., under review). There was a significant difference in the positive interaction (measured with the DRS) between the CST-PD and TAU arms at follow-up with the original data [F(1,40) = 4.574, p = 0.039, mean difference = 1.72 95% CI = 0.09, 3.34, p = 0.039] and with imputed data (mean pooled difference = 1.24, p-value varied between 0.021 and 0.131).

ANCOVA revealed no significant difference in burden (as measured with the ZBI) between the CST-PD and TAU groups at follow-up with the original data [F(1,39) = 2.379, p = 0.131, mean difference = -3.70, 95% CI = -8.55, 1.15, p = 0.131] or with imputed data (mean pooled difference = -2.44, p-value varied between 0.189-0.415). There was no significant difference in mental health (as measured by the SF-12 mental health sub-scale) between the randomisation arms at follow-up with the original data [F(1,42) = 0.083, p = 0.775, mean difference = -0.75, 95% CI = -5.98,

4.48, p = 0.775] or with imputed data (mean pooled difference = 0.65, p-value varied between 0.606 – 0.927).

Quality of life was measured with EQ-5D index score and EQ-5D visual analogue scale, which revealed no significant differences between CST-PD and TAU groups with original data [EQ-5D index with outliers: F(1,44) = 3.367, p = 0.073, mean difference = 0.09, 95% CI = -0.01, 0.19, p = 0.073; EQ-5D index without outliers: F(1,42) = 0.712, p = 0.403, mean difference = 0.03, 95% CI = -0.04, 0.10, p = 0.403; EQ-5D VAS: F(1,43) = 0.767, p =0.386, mean difference = 3.70, 95% CI = -4.82, 12.22, p = 0.386]. When multiple imputation was applied, EQ-5D index including the outliers was significant for the second and sixth imputation (mean pooled difference = 0.05, p-value varied between 0.025 – 0.960); however, when the two outliers were removed, EQ-5D was not significant between CST-PD and TAU groups (mean pooled difference = 0.001, p-value varied between 0.460 – 0.976). The EQ-5D VAS was not statistically different between CST-PD and TAU groups at follow-up (mean pooled difference = 2.03, p-value varied between 0.476 – 0.705). Therefore, there was no statistically significant difference in quality of life (as measured by EQ-5D index and EQ-5D VAS) between the two randomisation arms.

Table 9.3 Unadjusted means for each outcome measure for CST-PD and TAU at baseline and follow-up (n = 57 life partners).

	CST-PD				TAU			
	Baseline		Follow-up		Baseline		Follow-up	
Measure	N	mean (SD); range	N	mean (SD); range	N	mean (SD); range	N	mean (SD); range
RSS	30	24.80 (12.82); 0-42	21	28.0 (11.91); 1-42	27	30.19 (9.69); 10-42	25	28.5 (7.97); 11-41
DRS positive interaction	30	8.8 (2.87);4-15	22	9.6 (2.77); 5-16	23	9.0 (3.78); 3-18	24	8.3 (2.74); 4-15
ZBI	25	33.4 (16.29); 3-62	20	32.7 (15.89); 7-65	27	29.1 (14.96); 2-74	24	32.3 (15.44); 5-69
SF-12 mental health	29	47.71 (11.38); 17.56-61.78	21	48.85 (11.09); 27.42-64.12	26	48.11 (10.83); 19.81-62.63	25	48.21 (12.55); 22.66-64.88
EQ-5D index *(with outliers)*	30	0.768 (0.26); -0.016-1.000	22	0.814 (0.14); 0.620-1.000	27	0.825 (0.20); 0.157-1.000	25	0.771 (0.23); 0.082-1.000
EQ-5D index *(without two outliers)*							23	0.827 (0.13); 0.620-1.000
EQ-5D VAS	30	72.7 (17.16);35-100	22	74.3 (17.94); 35-100	26	77.4 (16.45); 40-100	25	74.8 (19.66); 30-100

Abbreviations: CST-PD – Cognitive Stimulation Therapy in Parkinson's-related dementia; DRS – Dyadic Relationship Scale; EQ-5D – EuroQoL; EQ-5D VAS – EuroQoL visual analogue scale; RSS – Relationship Satisfaction Scale; SD – standard deviation; SF-12 – Short Form Health Questionnaire; TAU – treatment as usual; ZBI – Zarit Burden Interview.

Table 9.4 Estimated marginal means, standard errors and 95% confidence intervals (CI), and mean differences (95% CI and p-values) comparing CST-PD and TAU at follow-up (adjusted for baseline outcome variables) with original (non-imputed) data and with seven imputations (pooled results and the minimum and maximum p-values among each imputation are presented) (n = 57 life partners).

	Original data								7 imputations			
		CST-PD		TAU					CST-PD (n = 30)	TAU (n = 27)		
Measure	**n**	**mean (SE); 95% CI**	**N**	**mean (SE); 95% CI**	**MD**	**95% CI of MD**	**p-value**	**F-statistic**	**Mean (SE); 95% CI**	**Mean (SE); 95% CI**	**MD**	**p-value**
RSS	21	30.07 (1.30); 27.44,32.69	25	26.78 (1.19); 24.38,29.18	3.28	-0.32, 0.69	0.073	3.375	29.37 (1.26); 26.90,31.84	27.02 (1.32); 24.44, 29.60	2.35	Ns (0.082-0.308)
DRS positive interaction	22	9.63 (0.56); 8.50, 10.76	21	7.91 (0.57); 6.75, 9.07	**1.72***	0.09, 3.34	**0.039**	4.574	9.40 (0.47); 8.57,10.42	8.30 (0.50); 7.28,9.23	1.24	3rd imputation significant (0.021-0.131)
ZBI	18	30.39 (1.80); 26.74, 34.04	24	34.09 (1.56); 30.93, 37.24	-3.70	-8.55, 1.15	0.131	2.379	31.41 (1.68); 28.12, 34.71	33.85 (1.72); 30.48, 37.22	-2.44	Ns (0.189-0.415)
SF-12 mental health	21	48.49 (1.89); 44.67, 52.30	24	49.24 (1.77); 45.66, 52.81	-0.75	-5.98, 4.48	0.775	0.083	48.80 (1.71); 45.45, 52.16	48.15 (1.75); 44.73, 51.57	0.65	Ns (0.606-0.927)
EQ-5D index *(with outliers)*	22	0.84 (0.04); 0.77, 0.91	25	0.75 (0.03); 0.68, 0.82	0.09	-0.01, 0.19	0.073	3.367	0.81 (0.05); 0.70, 0.91	0.75 (0.04); 0.67, 0.83	0.05	2nd & 6th imputation significant (0.025-0.960)
EQ-5D index *(without two outliers)*	22	0.84 (0.02); 0.79, 0.88	23	0.81 (0.02); 0.76, 0.85	0.03	-0.04, 0.10	0.403	0.712	0.82 (0.03); 0.76, 0.87	0.82 (0.03); 0.76,0.87	0.001	Ns (0.460-0.976)
EQ-5D VAS	22	77.00 (3.02); 70.90, 83.09	24	73.30 (2.89); 67.47, 79.13	3.70	-4.82, 12.22	0.386	0.767	75.57 (2.63); 70.42, 80.73	73.54 (2.75); 68.15, 78.94	2.03	Ns (0.476-0.705)

Notes: * $p < 0.05$

Abbreviations: CI – confidence intervals; CST-PD – Cognitive Stimulation Therapy in Parkinson's-related dementia; DRS – Dyadic Relationship Scale; MD – mean difference; Ns – not significant; RSS – Relationship Satisfaction Scale; SE – standard error; TAU – treatment as usual; VAS – visual analogue scale; ZBI – Zarit Burden Interview.

9.4.5 Serious adverse events and adverse events

In the current study, six serious adverse events (SAE) occurred with five people with PRD (one participant had two SAEs) and none occurred with care partners. All SAEs were reported to the principal investigator and the chief investigator. Four occurred with participants in the CST-PD group and two in the TAU group. The reasons for serious adverse events were hospitalisation (n = 4), prolonged stay in the hospital (n = 1) and worsening health (n = 1). None of the SAEs was a death and none was deemed to be related to the trial. Three dyads withdrew from the trial as a result of the SAE (two in the therapy arm and one in control arm).

A total of 70 adverse events (9 research incidents, 18 protocol violations and 43 protocol deviations) were reported in weekly phone calls with dyads. The majority of the adverse events were related to the health of one member of the dyad (i.e. feeling poorly, developing a cold or infection, having a fall, change in medication, etc). There was a relatively equal number of research incidents (four in CST-PD and five in TAU), protocol violations (nine in CST-PD and nine in TAU) and protocol deviations (20 in CST-PD and 23 in TAU) between the two arms. None of the adverse events were due to dyads' participation in the trial.

9.5 Discussion

9.5.1 General discussion

This study was a secondary analysis of a pilot feasibility single-blind two-arm randomised controlled trial of CST-PD exploring relationship satisfaction, burden and well-being in life partners of people with Parkinson's-related dementia. To date, there are a limited number of psychosocial interventions undertaken with people with PRD and their care partners (Connors et al., 2018; Farzana et al., 2015; Hindle et al., 2013, 2018); therefore, CST-PD is potentially an important contribution to widening the evidence-base for psychosocial therapies in complex types of dementia.

The current study revealed that CST-PD was not superior in regards to improving relationship satisfaction, burden, mental health and quality of life of life partners, which could be due to numerous reasons. The sample size in the study was small and it lacked power in order to detect change as the power calculations revealed that the required sample size is 128 participants. On the other hand, this is one of the few psychosocial intervention studies in this population and adds to the knowledge-base regarding dyadic interventions in complex types of dementia. It is noteworthy that the attrition rate was higher in the CST-PD group (14%) compared to the TAU arm (3.5%) making the two groups imbalanced, but importantly, two dyads decided to discontinue participation in the therapy due to difficulty in engaging in conversations and finding the therapy somewhat challenging for the person with PRD. This raises an important question whether the intervention is suitable for every dyad and whether it needs further tailoring and personalisation. Few dyads in the INVEST trial saw the therapy as a chore and an additional thing to do on top of already existing care-related responsibilities (McCormick et al., in press) which could explain the non-significant findings in emotional, psychological and relationship aspects in the current study. In the INVEST study, however, the care partners' burden, stress, quality of life and relationship satisfaction had improved (Leroi et al., under review) which could be due to a higher sample size (i.e. 76 dyads).

One significant finding emerged from this study. In the CST-PD arm, positive interactions between partners increased, which could potentially be explained by multiple interrelated factors: (1) the therapy provided dyads with an opportunity to communicate, reminisce and spend quality time together which the dyads might not have been doing much on a daily basis, (2) the life partners took a break from care-related tasks and responsibilities and directed their focus onto more positive aspects and emotions, (3) the dyad may have seen or felt themselves again as a couple rather than in the roles of 'caregiver' and 'care-recipient', and (4) including family members and friends in the therapy process allowed for socialising and strengthening relationships with significant others. All of these reasons are

hypothetical and were not tested specifically in the current study but they are questions of interest and should therefore be explored in more detail in the future.

The primary objective in the INVEST study, from which Study 5 stems from, was to explore feasibility, tolerability and acceptability of the intervention and as such was not specifically designed to improve or alter relationship satisfaction or quality. It is possible that life partners who were delivering the therapy may have taken on an additional role of the therapist which could have influenced how they felt about their dyadic relationship with the care recipient. As demonstrated by the first four studies in this PhD, the relationship dynamics within PRD have already changed and it may be difficult to improve relationships and restore premorbid relationship satisfaction, particularly due to the progressive and complex nature of PRD. Notwithstanding the non-significant findings in this study, focusing on strengthening relationships is important as strong relationships can lengthen lifespan (Mineo, 2017), contribute to happiness (Hassebrauck & Fehr, 2002; Russell & Wells, 1994) and affect life satisfaction (Falconier et al., 2015; Heller et al., 2004), remaining a fruitful avenue for future research.

9.5.2 Methodological strengths

The current study had multiple strengths. The INVEST study employed a randomised design with two arms (experimental and control) which were both active arms throughout the study and received a similar amount of contact with the research team (except for an additional therapy training visit in the CST-PD arm). The study recruited participants from multiple sites strengthening the generalisability of the trial findings across the recruiting sites in England. The INVEST study also recruited to target (76 dyads) of whom 57 were life partners, making it one of the largest feasibility studies with life partners of people with PRD. The inclusion of several scales allowed for multi-faceted exploration of life partners' mental and emotional health as well as their relationship with their partner. The outcome assessments were found to be feasible and acceptable to participants and could be considered as potential measures for the future definitive trial (McCormick

et al., in press). ANCOVA as a statistical analysis is considered a robust analysis that performs well even if data appears to have non-normal distribution (Barrett, 2011; Rheinheimer & Penfield, 2001; Vickers, 2005). ANCOVA is also considered stronger than a non-parametric analysis (i.e. the Mann-Whitney test) as it produces the effect size between the two groups, whereas the Mann-Whitney test provides only the p-value (Vickers, 2005); the former is recommended by CONSORT guidelines (Moher et al., 2010).

The missing values were imputed with the multiple imputation method according to the intention-to-treat principle which analyses data according to the randomisation. Multiple imputation was chosen as it is considered to be one of the best methods in *'restoring the natural variability of the missing values […] which results in a valid statistical inference'* (Kang, 2013, p. 405) and works well even when sample sizes are low and data are non-normally distributed (Graham, 2009; Graham & Schafer, 1999; Kang, 2013). In the current study, the residuals of each variable in one of the two arms appeared to be non-normally distributed; however, the data were not transformed due to the non-interpretability of results with transformed values. Several other missing value replacement methods exist but these were not chosen for the following reasons. First, the deletion method and single imputation methods could introduce bias and reduce the statistical power of the study (Kang, 2013). Second, methods of single imputation (i.e. mean substitution, regression imputation and last observation carried forward) could lessen the variability of values and minimise standard errors whilst not adding any new information (Ali et al., 2011; Horton & Kleinman, 2007; Kang, 2013; Malhotra, 1987). And finally, advanced model-based strategies such as the maximum likelihood and expectation-maximization were not applied as they can bias the estimates and underrate the standard error (Kang, 2013); therefore, the multiple imputation method was selected.

Furthermore, the INVEST study can be considered a complex intervention according to the MRC framework (Craig et al., 2008) due to the interacting components, several groups (people with PRD and care partners), variability in terms of disease

(PD-MCI, PDD, DLB) and symptoms (motor, psychiatric, cognitive), and the therapy that was tested, which was specifically adapted for people with PRD. The process evaluation of the INVEST study demonstrated that the CST-PD intervention was well accepted, tolerated and feasible (McCormick et al., in press) showing it can be feasible for a full-scale RCT trial in the future.

9.5.3 Limitations

The limitations of the study should be acknowledged. First, although the INVEST study recruited to target, the current study included a relatively small sample size and the dyad number in the control arm was less than 30, which is lower than recommended by Whitehead and colleagues (2016). Due to the small sample size, comparisons between clinical syndromes (i.e. PD-MCI, PDD and DLB), between male and female life partners and amongst all recruiting sites were not possible but should be a future consideration with a larger sample size and appropriate power. Despite the small sample size, the current study is one of the largest studies evaluating a psychosocial therapy amongst life partners of people with PRD and is similar to the sample sizes reported in earlier pilot and feasibility studies in the UK (Billingham, Whitehead & Julious, 2013).

Second, the study applied the intention-to-treat principle; which on the one hand may be problematic over a longer period of time as people with neurodegenerative conditions deteriorate making predictions of disease progression for each individual difficult, but on the other hand it may be more applicable for longitudinal studies, whereas the INVEST study was relatively short with only two time-points.

Third, the blinded researchers correctly guessed approximately half of the randomisation results, which may raise questions as to whether the assessors may have been unblinded to the randomisation allocation at any point throughout the dyads' participation in the study. A potential way to address unblinding includes monitoring and comparing researchers' answers after both baseline and follow-up visits, adding a measure of certainty of the researchers' randomisation guess and

having different researchers visiting the dyad at baseline and follow-up visits. In the INVEST study, the researchers that undertook the baseline assessment visits were the same at follow-up visits, when possible, as the dyad was familiar with the researchers from the first visit increasing rapport between the participants and researchers which is important as the assessment battery included sensitive measures.

Fourth, the research team lacked resources to monitor and observe the dyads' receipt and delivery of the intervention and how many therapy sessions they had actually completed over the course of 12 weeks, so it is unknown whether a higher adherence to therapy would have an impact on the outcome results for the CST-PD arm. Finally, although most of the results were non-significant in the current study, it should be acknowledged that the power of the study was relatively low.

9.5.4 Future directions

The INVEST study was a feasibility study which tested whether the intervention was acceptable, well tolerated and feasible to conduct and whether recommendations would be made for a definite full-scale RCT trial. The current study is an important addition in expanding our understanding of psychosocial therapies for care partners of people with PRD. However, some recommendations are advised for a future study.

The subsequent study should recruit a larger cohort and include a higher proportion of people with PD-MCI, PDD and DLB and their male and female life partners to be able to make comparisons among clinical syndromes and life partners' gender. In terms of outcomes, a positive trend was noted towards improvement on the relationship satisfaction scale and the next study could apply a variety of assessments measuring relationship satisfaction, relationship quality, various types of intimacy and attachment style to determine what specifically changes as a result of doing a psychosocial intervention and what contributes to the change. Furthermore, the current study measured negative aspects related to care

provision (i.e. burden) but there is increasing evidence that care partners may also experience many positive feelings related to care provision (Yu, Cheng & Wang, 2018) which the subsequent study should assess. The next study could also take into account the specific factors of burden (Vatter et al., 2018b) and evaluate whether there might be an improvement in various aspects of burden following the intervention.

The future study could consider adding a telephone and/or tablet-based app where participants could record the therapy sessions and the reason why they could not do the therapy if they had planned to do it, and the app could send gentle reminders to do therapy if no sessions have been completed within a week. The CST-PD could also be potentially adapted and developed into an app which could include a diverse range of topics and games that are stimulating, engaging and enjoyable for the dyad, a storage space for music and own personal photos making the therapy more individualised, and an option to record and rate the session; however, developing an app-based intervention is costly and time-consuming and may take a few years to develop and pilot. Moreover, the blinding procedure, which is complex and challenging in non-pharmacological trials due to the high risk of becoming unblinded, should be monitored to potentially minimise bias and ensure blinded researchers do not become unblinded to the randomisation allocation.

9.6 Conclusion

There are currently a limited number of psychosocial interventions specifically adapted for people with more complex types of dementia such as PDD and DLB and their life partners. The preliminary findings demonstrated that CST-PD increases positive interactions with the person with PRD; however, improvements in other domains, such as relationship satisfaction, mental health, burden and quality of life in life partners, were not noted in the CST-PD group. The CST-PD makes a valuable contribution to expanding the evidence- and knowledge-base of well-accepted

psychosocial interventions for life partners of people with PRD and a future trial should be conducted with modifications based on these findings.

CHAPTER 10: Discussion

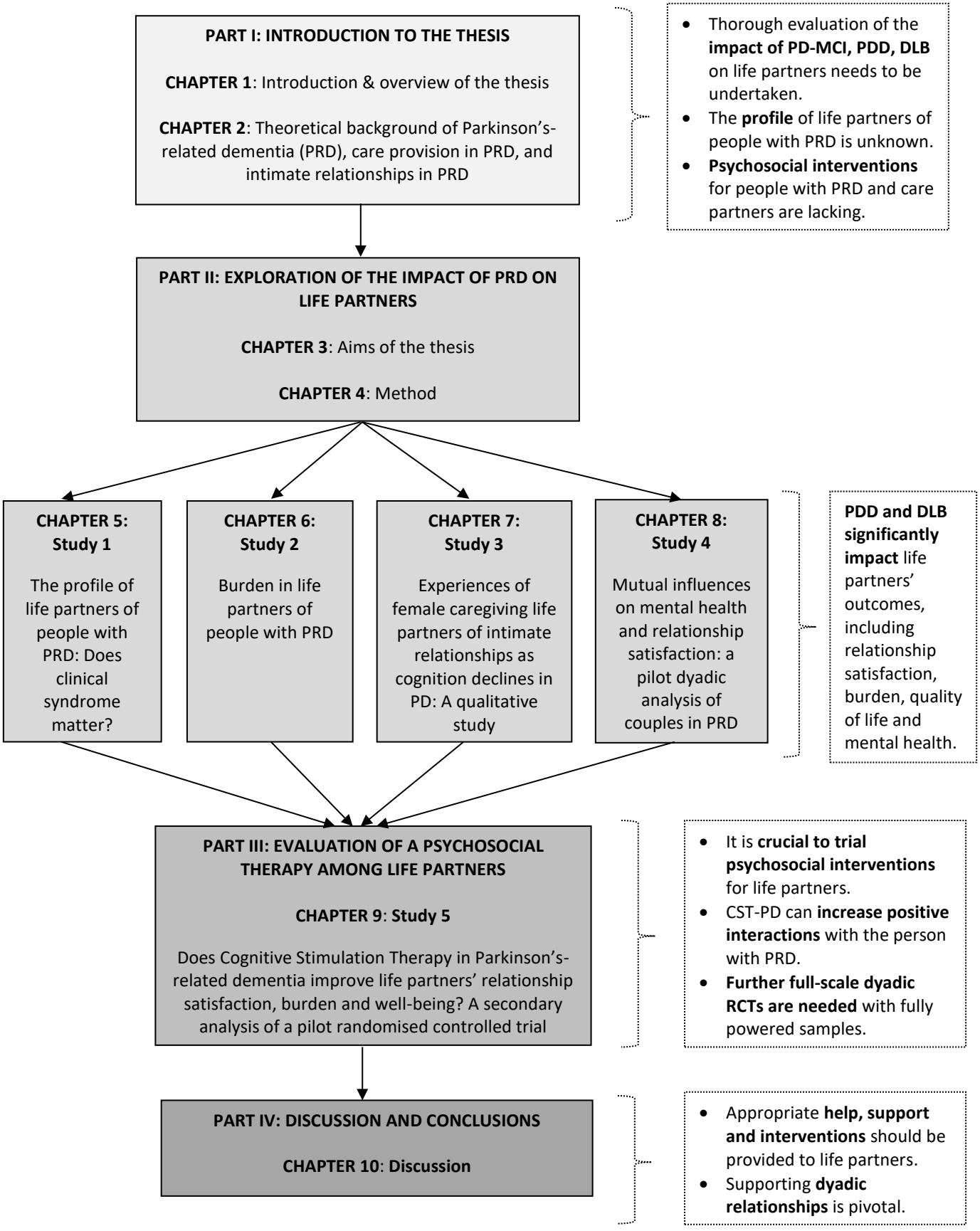
10.1 Summary of findings

In this set of studies, summarised in Figure 10.1, I have explored the profile of life partners of people with Parkinson's-related dementia (PRD), examined the impact of PRD on life partners' outcomes, and evaluated the effects of Cognitive Stimulation Therapy in Parkinson's-related dementia (CST-PD) on life partners. Specifically, my studies demonstrate that life partners of people with advanced cognitive impairment in PD, such as PDD and DLB, have poor levels of mental health and high rates of relationship dissatisfaction, burden, stress and negative feelings, including resentment, sadness, frustration and worry for the future. The qualitative study found that many life partners experience a role transition from spouse to care partner and feel that they have lost their own freedom and independence due to the progression of dementia in PD. At the same time, life partners cherish their marital vows and exhibit resilience, acceptance and adjustment despite the multiple challenges and complex nature of PRD.

My studies revealed that burden is experienced differently among life partners of people with PRD compared to other types of dementia. In deconstructing the underlying factors driving burden, two new factors emerged, namely 'interference with personal life' and 'concerns about future', highlighting that type of dementia may determine outcomes in life partners. This led me to evaluate the dyadic relationships of people with PRD and their life partners in more depth. For this, I chose the actor-partner interdependence model (APIM) which demonstrated that health-related outcomes, such as anxiety, depression and quality of life, are closely linked to relationship satisfaction. This suggests that these constructs should be taken into consideration in future dyadic intervention studies. Following the four exploratory studies examining the impact of PRD on life partners, I evaluated the effects of the pilot randomised controlled trial (RCT) of individualised Cognitive Stimulation Therapy adapted for PRD (CST-PD) on life partners. This final study demonstrated that CST-PD may increase positive interactions with the person with

PRD. Given the paucity of evidence-based psychosocial interventions for people with PRD and their life partners, CST-PD make a valuable contribution to the field and highlights the need for fully powered psychosocial interventions to assess efficacy.

Figure 10.1 Summary of the PhD studies and findings.



10.2 Contribution of the thesis

- ***The findings within my thesis add new knowledge regarding life partners of people with Parkinson's-related dementia.*** Study 1 is the first study to outline the profile of caregiving life partners of people with PD-MCI, PDD and DLB collectively. The examination of the factor structure of the Zarit Burden Interview among life partners of people with PRD, illustrated in Study 2, is new. The qualitative study describes the experiences of life partners which has not been done before for PD-MCI, PDD and DLB collectively (Study 3), although the experiences of care partners of people with PD, PD-MCI and PDD were evaluated in a recent study (Lawson et al., 2018). Study 4 investigates mutual influences in health-related outcomes and relationship satisfaction within PRD through dyadic analysis which is novel and adds to our knowledge regarding reciprocal effects in couples. Finally, Study 5 is innovative as it evaluates the effects of Parkinson's-adapted Cognitive Stimulation Therapy on life partners which has not been trialled before.

- ***The results of my thesis support and extend previous research with life partners of people with neurodegenerative conditions.*** A large proportion of caregiving life partners of people with PD, PD-MCI, PDD, DLB as well as AD, vascular dementia and frontotemporal dementia exhibit burden, stress, and lower quality of life and mental health (Brodaty & Donkin, 2009; Cheng, 2017; Ory, Hoffman, Yee, Tennstedt, & Schulz, 1999; Pinquart & Sörensen, 2003a). This demonstrates that a neurodegenerative condition in an individual can significantly affect the care partner, often more so than other conditions. However, the studies in this thesis showed that the impact of PRD on life partners differs when compared to the non-PD types of dementia. Comparative studies of neurodegenerative conditions consistently show that care partners of people with PDD and DLB have worse outcomes compared to other types of dementia, such as vascular dementia or AD (Davis et al., 2011; Lee et al., 2013; Ricci et al., 2009; Roland & Chappell, 2017; Shin et al., 2012; Svendsboe et al., 2016; Wu et al., 2018). This thesis also found that caregiving life partners of people with PRD provide more hours of care each day compared to non-PD type dementia (Brodaty & Donkin, 2009) and to the care

partners in the UK (NHS Digital, 2017). These differences may be due to the collective impact of motor, psychiatric and cognitive symptoms of PRD highlighting the unique, multifaceted and complex nature of PRD. Thus, it was important to focus on life partners of people with PD-MCI, PDD and DLB to understand how, and to what extent, PRD impacted their life and well-being.

Study 1 found differences in the outcomes of life partners according to the clinical syndrome of the care recipient. Life partners of people with PDD and DLB have higher rates of burden, depression and feelings of resentment compared to life partners of people with PD-MCI. In addition, higher stress levels, lower relationship satisfaction and fewer positive interactions with the care recipient are common among life partners of people with PDD, whereas higher anxiety levels and poorer mental health are more prominent among life partners of people with DLB, in comparison to PD-MCI stage. This agrees with findings from previous studies which have shown that once cognitive impairment emerges in PD, burden (Cifu et al., 2006; Jones et al., 2017; Leroi et al., 2012a; Martinez-Martin et al., 2015; Szeto et al., 2016), strain (Carter et al., 2008) and stress (Aarsland et al., 2007; Lawson et al., 2018) are higher, and quality of life is lower (Lawson et al., 2018) among life partners. One explanation for these differences could be the duration of PD and cognitive impairment as PDD develops gradually over a number of years giving life partners more time to get accustomed to the condition, adjust to the changes and seek possible treatment opportunities. At the same time, the duration of care provision in PDD is longer and thus, life partners may have experienced a role transition from spousal/life partner role to a care partner/carer role changing the interaction and dyadic relationship of the couple. In contrast, DLB develops and progresses more rapidly than cognitive impairment in PD and the changes may be much more unexpected and sudden which can increase anxiety, fear and worry among life partners, subsequently affecting their mental health and quality of life. Therefore, it can be concluded that the specific clinical syndrome in PD plays an important role in determining life partner outcomes. In addition, Study 1 described the profile of life partners of people with PRD and highlighted that some life partners may be at risk of developing poor health outcomes. Understanding the

typical profile of caregiving life partners of people with PRD may facilitate the identification of caregiving partners in the community and during the appointments with health specialists who could potentially provide help and support to life partners in order to prevent their burnout.

The demands of continuous care provision in PRD place an immense strain on life partners and increase burden. With the progression of cognitive impairment in PD, the burden in life partners also increases as seen in Study 2, in line with earlier research (Grün et al., 2016; Leroi et al., 2012a; Martinez-Martin et al., 2015; Mosley et al., 2017). Since the outcomes of life partners of people with PDD and DLB are worse compared to those life partners caring for people with other types of dementia, it was important to investigate how life partners experienced burden. The most widely used burden scale in PRD (Leiknes et al., 2015), the Zarit Burden Interview (ZBI; Zarit et al., 1980), was examined to identify the factor structure. Unlike earlier studies in dementia, which demonstrated that the ZBI was three-dimensional (Ankri et al., 2005; Leggett et al., 2011; Pillemer et al., 2018; Smith et al., 2018; Springate & Tremont, 2014), a five-factor structure of the ZBI scale emerged among life partners of people with PRD. The two new factors, 'interference with personal life' and 'concerns about future', could be considered in the context of the qualitative study showing that, as a result of care provision, life partners' own time and freedom are reduced leaving them with less time for themselves and for interacting with others. Similarly, in order to be able to provide care, life partners have to be well enough but most life partners are also older adults themselves and may have physical and/or mental health ailments decreasing their ability to provide care (Berger et al., 2017; Tan et al., 2012). This, in turn, increases uncertainty and fear among life partners with respect to whether they can continue providing care but if they cannot, the worry regarding who will look after the care recipient increases. It may be advantageous for a future intervention to target these specific types of burden among life partners that could ease and reduce stress, overall burden, negative emotions and burnout.

- ***My thesis has highlighted a core component of the caregiving relationship.*** People have a strong desire to be loved and to belong, which motivates them to find romantic partners. The "belongingness hypothesis" outlines that we need to create and preserve a minimum quantity of social interactions with others, which are positive, pleasant, meaningful, and stable (Baumeister & Leary, 1995). Thus, many people yearn to find intimate partners and establish long-term relationships. Having good relationships is crucial because relationships can determine happiness as well as significantly impact the physical and mental health of people, as determined by one of the biggest longitudinal studies spanning 75 years, "The Harvard Study of Adult Development" (Mineo, 2017) and by several cross-sectional studies (Bookwala & Franks, 2005; Kiecolt-Glaser & Newton, 2001; Waldinger & Schulz, 2010). Equally, reduced relationship quality and relationship dissatisfaction can contribute to poor mental health outcomes, such as depression (Baumeister & Leary, 1995; Beach et al., 1985; Beach et al., 2003; Carr et al., 2014; Clare et al., 2012; Levenson et al., 1993; Proulx et al., 2007). Researchers have determined that good relationships prolong lifespan (Mineo, 2017), determine life satisfaction (Falconier et al., 2015; Heller et al., 2004) and happiness (Hassebrauck & Fehr, 2002; Russell & Wells, 1994), despite the daily challenges of managing physical health issues (Waldinger & Schulz, 2010). Therefore, having good relationship quality is valuable as it can positively influence our lives. Many couples in my PhD studies have been together or married for over 40 years but the neurodegenerative condition of one partner has significantly disrupted the relationships and brought about many unexpected changes, including loss of a relationship, intimacy, partner, connectedness, closeness and interaction, as evidenced in the qualitative study (Study 3) and in earlier studies in dementia (Boylstein & Hayes, 2012; Evans & Lee, 2014; Pozzebon et al., 2016).

- ***This thesis has made an important contribution through a detailed exploration of the experiences of spouses and life partners of people with PD-MCI, PDD and DLB, thus adding to existing knowledge.*** The majority of life partners interviewed in Study 3 experienced a reduction in relationship satisfaction and intimacy as a result of the PRD of the care recipient. Life partners are more emotionally distanced from

the person with PRD despite spending more time together. One of the main reasons why relationship satisfaction alters is the diminished ability of the person with PRD to communicate resulting in poorer communication between the couple. Although many couples talk and interact, several couples no longer have a meaningful conversation, which they miss and find particularly difficult. This is in line with earlier research among life partners of people with dementia in which decreased communication has been regarded as one of the most challenging aspect for life partners resulting in relationship dissatisfaction (Boylstein & Hayes, 2012; Clare et al., 2012; de Vugt et al., 2003; Evans & Lee, 2014; Garand et al., 2007; Pozzebon et al., 2016). The lower levels of relationship satisfaction due to lack of conversation, communication and interaction are somewhat predictable as they can act as a barrier to closeness, mutuality, reciprocity, intimacy and bond in the couple. Many of these components are fundamental to a relationship and their absence can lead to less happiness, worse outcomes and shorter life duration for both partners. This has been demonstrated by the longitudinal Harvard Adult Development Study (Mineo, 2017; Waldinger & Schulz, 2010). My findings from Study 3 suggest that future interventions should focus on increasing socialising and interaction between people with PRD and their life partners. Targeting communication could subsequently improve the relationship quality and satisfaction for both partners.

- ***My work has extended our understanding of the reciprocal impacts of life partners and individuals with a chronic neurodegenerative condition.*** Earlier studies with dyads of people with PRD and life partners have explored how the motor, psychiatric and cognitive symptoms of people with PRD are associated with or predict the outcomes of life partners (Aarsland et al., 1999a; Lawson et al., 2017; Leroi et al., 2012a; Martinez-Martin et al., 2008; Roland et al., 2010; Tan et al., 2012). However, most studies have not taken into consideration how the outcomes of each person can mutually influence their own outcomes as well as the outcomes of their partner jointly. The Study 4 finding that anxiety scores of the person with PRD can contribute towards their own relationship dissatisfaction and towards relationship satisfaction in the life partners can be explained by a feedback cycle.

Although the sample size of the study was small, the results suggest that the mental health of the person with PRD can affect the relationship satisfaction of both partners highlighting that conducting dyadic analyses can be valuable and enrich our understanding of how partners influence each other. Such knowledge may not be gained if the outcomes of each partner are not studied together. The study also indicates that as the outcomes of people with PRD are linked to the outcomes of life partners, the dyadic interventions are important to consider which could potentially be advantageous for both partners or benefit one person who, in turn, could positively affect the other partner. Study 4 was an exploratory pilot study and the findings form the necessary first step to design a subsequent study in a larger sample using comprehensive measures of mental health and relationship satisfaction.

- ***My findings have corroborated the strong association between relationship satisfaction and burden.*** Previous studies in PD have shown similar results (i.e. Schrag et al., 2006), which adds confidence to my conclusions. This aligns well with the theoretical framework of this thesis, the adapted Stress-Appraisal model (Greenwell et al., 2015), whereby the cognitive and neuropsychiatric symptoms of the person with PRD are the primary stressors, necessitating help from a care partner (primary appraisal). Care partners' ability to cope with the caring role (secondary appraisal) can determine the outcomes of life partners, such as burden, stress and negative emotions (tertiary appraisal), but this model is mediated by the quality of the dyadic relationship, which can possibly prevent negative outcomes for life partners (Goldsworthy & Knowles, 2008; Greenwell et al., 2015), particularly if the relationship quality is good. This could be due to how life partners view their role: if life partners see care provision as a natural progression of their relationship, it can be protective against negative outcomes (Gaugler et al., 2002; Gillies, 2011; Lawn & McMahon, 2014; Martin, 2016; Molyneaux et al., 2011b). Hence, supporting dyadic relationships is key in order to maintain better outcomes and well-being of life partners for as long as possible.

- ***My thesis has investigated an important question with clear clinical relevance.*** Researching relationships in older adults within PRD is important for a number of reasons. Looking at couples' relationships in PRD provides an opportunity to explore the hurdles, barriers and obstacles that couples face, what they find the hardest to accept and adjust to, and how they cope or deal with these issues and challenges. By gaining an insight into the day-to-day life of couples, we can understand better what help and support to provide in order to improve outcomes for both members of the dyad. The studies here demonstrate that neuropsychiatric symptoms and cognitive impairment of the person with PRD are the hardest to cope with, manage and accept, and it is these symptoms that contribute to more negative outcomes in life partners. This suggests that there may be an important role for specifically adapted psychosocial interventions that target cognitive impairment, communication and neuropsychiatric symptoms of the person with PRD. Preliminary results from the pilot CST-PD intervention (Study 5) found that as a result of participating in the therapy, the amount of positive interactions between the partners increased. This highlights that a dyadic therapy, which involves communication and interaction, can be beneficial for couples. The reasons why positive interactions increased could be potentially due to the dyad spending more time together, doing a meaningful activity, reminiscing, having a conversation and reciprocally listening to each other. In addition, life partners may have exhibited more patience, empathy and acceptance of the person with PRD, who may have appreciated greater empathy and patience, even if they may have found the therapy somewhat challenging. Life partners may have also accepted and acknowledged the situation they were in and started to prepare themselves for the upcoming future. All of these aspects emphasise that care provision may also have a positive side in terms of engaging with the care recipient, doing activities together and recognising the changes, which is evident from earlier studies (Yu et al., 2018). Understanding why positive interactions increased among those who undertook the CST-PD intervention remains a fruitful area for research.

- ***This set of studies has clear relevance for current care provision for people with PRD in the UK.*** Recently, changes have been proposed to NHS care practice

whereby local Clinical Commissioning Groups suggested closure of local hospital beds and reliance on 'Care at Home' and 'Dementia Rapid Response Team' services (Ball, 2017). These changes may increase burden among life partners as they have to be willing, available and well enough to take care of people with PRD at any time, which may not always be in their control. Whilst it is recognised that care partners are pivotal in the healthcare system (Carers UK, 2018; Department of Health and Social Care, 2018), it is important to take into account the needs of the life partner. Education, support, assistance and targeted psychosocial interventions that help to build resilience, increase coping and reduce burden, stress and chance of burnout in life partners are required.

10.3 Strengths of the thesis

A key strength of the PhD studies is the novelty of including life partners of people with three clinical syndromes within PD collectively, namely PD-MCI, PDD and DLB. Differentiating the stages adds to our understanding regarding the impact of these conditions on life partners. Earlier research has compared the outcomes of life partners of people with PDD and DLB to other types of dementia but has mostly excluded the PD-MCI group. Including the PD-MCI group is valuable because, as studies within this thesis highlight, life partners of people with PD-MCI also experience changes in their relationship and have increased negative feelings. Moreover, the umbrella term 'Lewy body spectrum disorders' includes PD-MCI, alongside PDD and DLB, and PD-MCI is a well-established harbinger to dementia. Thus, it is important to examine the pre-dementia stage in PD to fully explore the progression, transition and impact on outcomes of life partners and people with PRD as well as on their intimate relationship so that we can offer targeted support earlier.

Study 1, which explored the demographic and clinical profile of life partners, is the largest study to date of life partners of people with PRD. Similarly, Study 2, the factor analysis of the ZBI has an appropriate sample size (i.e. above 100 participants; Gorsuch, 1983; Kline, 1994) and the factors that emerged from the

analysis explained over 60% of the total variance which is considered acceptable for a scale to be valid (Hair, Black, Babin, Anderson, & Tatham, 2005). Study 5, which conducted a secondary analysis of a pilot RCT of the CST-PD, was one of the largest psychosocial intervention studies with life partners of people with PRD.

Another key strength of this PhD is the application of different analysis methods, which were chosen to answer specific research questions as well as the exploration of a wide variety of measures of life partners, which allowed a thorough and multifaceted examination of the physical and mental health of life partners, quality of life, relationship aspects, burden, stress and resilience. Study 1 evaluated the psychometric properties of the scales which is infrequently reported by many studies and this study contributed to the transparency and clarity of the research findings. Additionally, Study 3 employed a qualitative analysis where subjective opinions and views of life partners were extracted and in-depth information obtained (Anderson, 2010). A frequency of quotes table, generated in Study 3, determined and summarised the most important areas for life partners quantitatively. These were 'Emotional distance', 'Negative repercussions', and 'Motor and non-motor manifestations of PD'. In Study 4, the actor-partner interdependence model (APIM) was conducted with life partner-people with PRD couples finding mutual influences between the outcomes of life partners and the outcomes of people with PRD (i.e. actor and partner effects) and demonstrating that the outcomes of both members of the dyad are interconnected. Study 4 also reported the associations between health-related outcomes and relationship satisfaction in both partners within PRD which has not been done previously.

Finally, there are several strengths in Study 5, which evaluated the CST-PD intervention among life partners. The study is a complex intervention (Craig et al., 2008) for the following reasons: (1) the intervention was trialled with person with PRD-life partner dyads, (2) the people with PRD had a diagnosis of PD-MCI, PDD or DLB and varied in terms of their symptom presentation, and (3) the tested therapy was newly adapted and had not been trialled before among people with PRD. This study is also a RCT where participants had an equal chance of being allocated to the

treatment group or to the control group and RCT studies are generally considered gold-standard if conducted properly (Bothwell, Greene, Podolsky, & Jones, 2016). Recruitment took place in several sites in England which enhanced the value of the findings and reduced researcher bias as all participants were seen by different researchers. Taking the aforementioned aspects into account, Study 5 is an important addition to the field of psychosocial interventions for people with PRD and life partners.

10.4 Critical analyses of the thesis

Despite the multiple strengths of this thesis, several limitations of this thesis should be acknowledged. All studies, with the exception of Study 5, were cross-sectional and only obtained outcomes of life partners at one time-point, which precluded making long-term observations and exploring possible causality in relation to the studied outcomes among life partners. It may be useful to undertake a longitudinal study of the couples' relationships to observe and detect how relationships change over time and how couples cope with these changes, which may help with providing tailored and suitable interventions for couples as well as strategies to maintain the relationships.

While the sample size was considered appropriate for Studies 1, 2 and 3 (Gorsuch, 1983; Guest, Bunce, & Johnson, 2006; Kline, 1994), a larger sample in the PhD studies could have increased generalisability of the findings. Studies 4 and 5 are pilot exploratory studies; thus, power calculations were not conducted and the findings should be interpreted with caution. It was also not feasible to compare outcomes of life partners according to the diagnosis of the person with PRD (i.e. PD-MCI, PDD and DLB) in Studies 4 and 5 due to the small sample size. However, Study 4 had a bigger sample size than earlier studies with older adults (Mavandadi et al., 2014; Regan et al., 2014; Walker et al., 2013) and study 5 was one of the largest psychosocial interventions undertaken with life partners of people with PRD to date. Since life partners in all PhD studies were predominantly female, it was not possible to make comparisons between male and female life partners and the

gender imbalance limited the generalisability of the findings to a wider population of couples where one person has PRD.

The qualitative study only included white female life partners which prevented exploration of whether ethnicity, gender or relationship type (i.e. same-sex relationship, parent-child relationship) had an impact on the experiences of life partners. Moreover, certain life partners may be more willing to come forward to talk about their experiences and take part in research than other life partners and these participants may have a different experience to life partners in the seldom-heard groups. It would be valuable to include a heterogeneous sample with life partners of different cultural backgrounds, ethnicities and socioeconomic status in order to increase cross-cultural validity of the results. At the same time, the majority of care partners in the UK tend to be women (Alzheimer's UK, 2015; Carers UK, 2017) and there are more males with PD than women with PD (Van Den Eeden et al., 2003; Wooten, Currie, Bovbjerg, Lee, & Patrie, 2004), which is reflective of the sample that was recruited in the PhD studies.

The response rate in the postal questionnaire survey in Studies 1 and 2 could be considered low (43%) and it may have been advantageous to post out another copy of the questionnaire to potential participants, phone participants if they had not returned the questionnaire within a few weeks, and consider providing small monetary incentives. Moreover, it was not possible to obtain data on motor, neuropsychiatric and cognitive symptoms of people in PRD in postal questionnaires which lessened the sample size for disease-related variables in Studies 1 and 2. As data for life partners were extracted from the INVEST study for Studies 1 and 2, a sample selection bias was present. Studies 1 and 2 also had a self-selection bias as participants who took part in the postal questionnaire study made a decision whether to participate in the study or not. These biases could have been overcome by applying only one recruitment method for the studies. However, as the data in Studies 1 and 2 were amalgamated from the INVEST study and the postal questionnaire study it was not considered a major limitation.

All measures in the study were self-reported by life partners who provided a subjective account of their health and care provision-related aspects which were largely dependent on their care-related responsibilities, amount of stress and burden they experienced, reactions and abilities to cope and to bounce back in stressful situations (i.e. resilience) at the time of completing the assessments. The PhD studies did not explore the health history of life partners in detail which could have provided a more thorough understanding of the mental and physical health needs of life partners. Furthermore, although the measures of people with PRD were completed with blinded assessors in the INVEST study, they were largely self-reported by people with PRD who may have provided an inaccurate account of the symptoms they experienced due to cognitive impairment. It is also noteworthy that the motor, cognitive and psychiatric symptoms of PD-MCI, PDD and DLB are highly individual, may fluctuate daily or even hourly and may deteriorate in a few months or years; hence, it was only possible to get a glimpse of the symptoms at one time-point and not observe the symptom fluctuation over time, which is important to take into account in future research.

Missing data occurred in all quantitative studies both within life partner and person with PRD outcome measures, whereby either single items or entire scales were missing. The reasons for missing data were varied: data were missing at random, person with PRD may have been unable to complete the assessments due to fatigue and/or cognitive impairment, a few dyads had withdrawn from the study and some data were missing not at random (i.e. the participant could not complete a scale containing sensitive information in the presence of a partner). The missing data were addressed with expectation-maximization method in Study 1 and with multiple imputation method in Study 5. In Study 2 the missing data were removed listwise. However, in Study 4 the missing data were not addressed as the available options of handling missing data (such as multiple imputation) may pose challenges and obstacles for multilevel models with interaction effects (Grund, Lüdtke, & Robitzsch, 2018) and one needs to be cautious of imputing missing data due to the nested data. Therefore, the risk of conducting erroneous analyses is high and it was decided that the data variables in Study 4 will not be imputed. Notwithstanding the

decision of not imputing the missing data in Study 4, tackling missing data remains important because missing data that is not addressed can lead to a considerable reduction of the original recruited sample size which subsequently decreases power and precision (Sterne et al., 2009).

In Study 5, the CST-PD arm had a higher attrition rate than the TAU arm which may raise questions regarding the tolerability of the intervention. Furthermore, it was not possible to observe and monitor adherence, i.e. the amount of CST-PD sessions delivered and received among participant-dyads, which limits the understanding whether dyads adhered to the therapy protocol and completed the recommended amount of therapy sessions (i.e. 20 to 30 minute sessions, 2-3 times per week for 12 weeks).

10.5 Recommendations for future research

One of the main recommendations for a future study is applying a longitudinal design which could support the accuracy of the findings and lead to a better understanding of the cause and effect of the studied variables. Employing a longitudinal design could also help to monitor how relationships and outcomes of life partners change and transform over time, particularly as PRD deteriorates, and explore what impacts and predicts relationships and outcomes, which would depict a better understanding of the long-term impact of PRD on life partners. It would also be useful to examine the model fit of the ZBI among life partners between two or more time-points, which could provide a more accurate portrayal of the specific burden constructs.

Most PhD studies had a relatively low sample size and future studies could aim to recruit more life partners of people with PD-MCI, PDD and DLB which would make it possible to compare outcomes according to the clinical syndrome, gain a deeper understanding of the experiences of life partners and increase generalisability to a wider population (i.e. external validity). A subsequent study could diversify the sample by recruiting: (1) male and female life partners to compare their

experiences according to gender, (2) people with different cultural backgrounds and ethnicities to increase cross-cultural validity, and (3) people in distinct types of relationships, such as marital, long-term partnerships, same-sex partnerships, parent-adult child or family relationships to observe whether type of relationship with the care recipient can have a similar impact on the life partner. The qualitative interviews could be conducted with couples together which can result in rich and meaningful data providing the perspectives of both partners.

Several outcomes were explored in the PhD studies but a subsequent study could employ a comprehensive assessment battery, including disease-related measures for people with PRD and measures of positive aspects and experiences of care provision, coping mechanisms, attachment styles, social support, relationship quality, mutuality and various types of intimacies for life partners. Future studies could also employ mixed methods where quantitative and qualitative methods are combined to portray a more accurate and complete picture of relationships in PRD. A systematic literature review is recommended to summarise the role and profile of care partners of people with PRD. One of the key aspects is also exploring how life partners cope with the changing care-related responsibilities and demands that the disease places on them and whether they apply additional coping strategies or have other coping mechanisms in place, which is important to take into consideration in future studies.

Future studies should continue exploring the feasibility and efficacy of psychosocial interventions among people with PRD and life partners to determine what psychosocial interventions are preferred by dyads and which are efficacious. By focusing on supporting dyadic interventions, it may be possible to preserve intimate relationships and delay institutionalisation of the person with PRD, which has cost saving implications and deep-rooted effects on both people with PRD and life partners.

Finally, a future study trialling a psychosocial intervention could be delivered in an app-format containing a range of topics and activities, storage for own music and

photos, and an option to video-record the sessions, so that the researchers could analyse the interactions of the dyad. There may also be scope to add an online support program for life partners in addition to doing the intervention with the care recipient, which has been found to be beneficial (McKechnie et al., 2014a). Another key suggestion is to provide additional information, education, training and tips to life partners to help with coping with day-to-day challenges which could alleviate burden, stress, negative feelings, emotions and experiences of care provision which subsequently can increase the ability of life partners to provide care for longer.

10.6 Implications for healthcare, policy and practice

The thorough examination of the outcomes of life partners of people with PRD demonstrated the major impact that PRD has on life partners and CST-PD showed that a specifically adapted psychosocial intervention can be beneficial for life partners in increasing positive dyadic interactions. My PhD highlights that clinicians, consultants, PD specialist nurses and other health care professionals should also focus on care partners to assess how they are coping, whether they need additional help and potentially provide some tailored support and interventions. Although this may be an extra resource for healthcare professionals in terms of time and offering specialised care, providing help and support to life partners will carry a positive effect for both members of the dyad and people with PRD may be cared by life partners for longer at their homes. The current research has implications for policy makers, stakeholders, clinicians as well as future clinical trials and practice as including life partners as active participants in psychosocial interventions and delivering dyadic interventions could potentially benefit both people with PRD as well as life partners.

Caregiving life partners are crucial in the healthcare teams as they provide continuous care to people with PRD, understand their needs and support them with daily activities of living. In the UK, informal care provision saves over £11 billion each year but government bodies and the healthcare system may not always recognise the value of life partners' support and the hardship of care provision as

the NHS is keen to reduce hospital stays of people with dementia meaning life partners have to provide care. In 2018, the Department of Health and Social Care and charities, such as Carers UK and Parkinson's UK, have published reports and actions plans for care partners encouraging health and social care services to ensure care partners are included, supported and valued, that care partners are recognised in the wider community and society, and that research focuses on seeking effective solutions to improve outcomes for care partners. This also corresponds to the Prime Minister's Challenge on Dementia 2020 which aims to make England the leading country in the world for dementia care, research and awareness (Department of Health, 2016). The aforementioned points are particularly important to consider for life partners of people with lesser known dementia such as PDD and DLB as life partners may often go unnoticed and not receive the support they need. The government, the NHS, charities as well as care partners should work closely together to support relationship-centred care and deliver appropriate help, support and interventions that benefit caregiving life partners.

10.7 Conclusions

The studies within this thesis highlight the profound effect that PRD can have on life partners, particularly due to psychiatric and cognitive symptoms of people with PRD which intensify with the progression of cognitive impairment in PD. Relationship dissatisfaction, burden, stress, negative feelings and poor mental health levels are common among life partners of people with PRD and for many life partners their life changes as a result of becoming a care partner. To date there is only minimal targeted support available for life partners of people with PRD and this thesis evidenced that a dyadic psychosocial intervention could benefit said individuals. As good relationships can be protective against negative outcomes in life partners, future studies should focus on supporting intimate relationships, take into consideration the physical, psychological, emotional and social needs of partners, and conduct fully powered psychosocial interventions that could benefit both people with PRD and life partners.

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Appendices

Appendix A: Ethical approval letter

NHS
Health Research Authority
Yorkshire & The Humber - Bradford Leeds Research Ethics Committee
Room 001
Jarrow Business Centre
Viking Industrial Park
Rolling Mill Road
Jarrow
NE32 3DT

18 January 2016

Dr Iracema Leroi
University of Manchester
3rd Floor, Jean McFarlane Building,
University Place, Oxford Road, Manchester.
M13 9PL

Dear Dr Leroi

Study title:	**A Psychosocial Therapy to Benefit People with Parkinson's-relatedDementia: A Feasibility and Exploratory Pilot Study of Individual Cognitive Stimulation Therapy (INVEST).**
REC reference:	**15/YH/0531**
IRAS project ID:	**166392**

Thank you for your letter of 10th January, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact the REC Manager, Katy Cassidy, nrescommittee.yorkandhumber-bradfordleeds@nhs.net.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Mental Capacity Act 2005

I confirm that the committee has approved this research project for the purposes of the Mental Capacity Act 2005. The committee is satisfied that the requirements of section 31 of the Act will be met in relation to research carried out as part of this project on, or in relation to, a person

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who lacks capacity to consent to taking part in the project.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for NHS permission for research is available in the Integrated Research Application System, www.hra.nhs.uk or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to contest the need for registration they should contact Catherine Blewett (catherineblewett@nhs.net), the HRA does not, however, expect exceptions to be made. Guidance on where to register is provided within IRAS.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS sites

The Committee has not yet completed any site-specific assessment (SSA) for the non-NHS research site(s) taking part in this study. The favourable opinion does not therefore apply to any non-NHS site at present. We will write to you again as soon as an SSA application(s) has been reviewed. In the meantime no study procedures should be initiated at non-NHS sites.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
Copies of advertisement materials for research participants [INVEST advert]	1.0	10 August 2015
Covering letter on headed paper [Covering letter]		
GP/consultant information sheets or letters [GP information letter]	1.0	10 August 2015
Interview schedules or topic guides for participants [Therapy guide]		
Interview schedules or topic guides for participants [Interview schedule - post intervention]		
IRAS Checklist XML [Checklist_11012016]		11 January 2016
Letter from funder [Letter from Funder]		
Non-validated questionnaire [ECQv3]		
Other [Demonstration of therapy manual adaptation]		
Other [Additional letters of support]		
Other [Role of consultee INFO]	1.0	10 August 2015
Other [Loss Capacity during study letter MAIN TRIAL]	1.0	10 August 2015
Other [SAE reporting form]	1.0	10 August 2015
Other [Randomisation letter - therapy]	1.0	
Other [Randomisation letter - no therapy]	1.0	
Other [Nominated consultee declaration FOOTAGE]	1.0	10 October 2015
Other [Personal Consultee declaration FOOTAGE]	1.0	10 October 2015
Other [Personal Consultee PIS FOOTAGE]	1.0	10 October 2015
Other [Nominated Consultee PIS FOOTAGE]	1.0	10 October 2015
Other [Personal Consultee PIS MAIN TRIAL]	1.1	06 January 2016
Other [Personal Consultee declaration MAIN TRIAL]	1.1	06 January 2016
Other [Nominated Consultee PIS MAIN TRIAL]	1.1	06 January 2016
Other [Nominated Consultee declaration MAIN TRIAL]	1.1	06 January 2016
Other [Loss Capacity during study declaration MAIN TRIAL]	1.1	06 January 2016

Participant consent form [Carer consent MAIN TRIAL]	1.0	10 August 2015
Participant consent form [2 WEEK FEASIBILITY Carer consent 10.10.2015]	1.0	10 October 2015
Participant consent form [2 WEEK FEASIBILITY Patient consent 10.10.2015]	1.0	10 October 2015
Participant consent form [Carer consent FOOTAGE]	1.0	10 October 2015
Participant consent form [Patient consent FOOTAGE]	1.0	10 October 2015
Participant consent form [Patient consent MAIN TRIAL]	1.1	06 January 2016
Participant consent form [Carer consent MAIN TRIAL]	1.1	06 January 2016
Participant information sheet (PIS) [Carer PIS MAIN TRIAL]	1.0	10 August 2015
Participant information sheet (PIS) [2 WEEK FEASIBILITY Carer PIS 10.10.2015]	1.0	10 October 2015
Participant information sheet (PIS) [2 WEEK FEASIBILITY Patient PIS 10.10.2015]	1.0	10 October 2015
Participant information sheet (PIS) [Carer PIS FOOTAGE]	1.0	10 October 2015
Participant information sheet (PIS) [Patient PIS FOOTAGE]	1.0	10 October 2015
Participant information sheet (PIS) [Carer PIS MAIN TRIAL]	1.1	06 January 2016
Participant information sheet (PIS) [Patient PIS MAIN TRIAL]	1.1	06 January 2016
REC Application Form [REC_Form_11112015]		11 November 2015
Referee's report or other scientific critique report [Review DeNDRoN]		
Referee's report or other scientific critique report [Internal review content]		
Referee's report or other scientific critique report [Review NCS]		
Referee's report or other scientific critique report [Review PD Trust]		
Research protocol or project proposal [Protocol 1.0 10.08.2015]	1.0	10 August 2015
Summary CV for Chief Investigator (CI) [CI CV]		
Summary CV for student [Student CV]		01 September 2015
Summary CV for supervisor (student research) [2nd Supervisor CV]		
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Flow chart of study]		
Validated questionnaire [ACE]	1.0	06 January 2016
Validated questionnaire [DCFS Lee]		
Validated questionnaire [NPI]		
Validated questionnaire [UPDRS and SE]		
Validated questionnaire [CSRI 1]		
Validated questionnaire [CSRI 2]		
Validated questionnaire [Remaining patient questionnaires]		
Validated questionnaire [Remaining caregiver questionnaires]		

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

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After ethical review

Reporting requirements

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:
http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

15/YH/0531	**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project.

Yours sincerely

pp

Dr Janet Holt
Chair

Email:nrescommittee.yorkandhumber-bradfordleeds@nhs.net

Enclosures: "After ethical review – guidance for researchers" *[SL-AR2]*

Copy to: *Ms. Julia Foster, Manchester Mental Health & Social Care Trust*

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Appendix B: Diagnostic criteria of PD, PD-MCI, PDD and DLB

Table B.1 Adapted (Fields, 2017) diagnostic criteria for PD (Gibb & Lees, 1988), non-motor symptoms of PD (Chaudhuri et al., 2011), PD-MCI (Litvan et al., 2012), PDD (Emre et al., 2007) and DLB (McKeith et al., 2017).

	PD (Gibb & Lees, 1988)	**PD-MCI** (Litvan et al., 2012)	**PDD** (Emre et al., 2007)	**DLB** (McKeith et al., 2017)
Core features required to diagnose	• Bradykinesia At least one of the following: • Muscular rigidity • 4-6 hertz rest tremor • Postural instability	• Diagnosis of PD (according to Brain Bank Criteria, Gibb & Lees, 1988) and gradual decline in cognitive ability in context of established PD. • Cognitive deficits on either formal neuropsychological testing or a global cognitive abilities scale. • Cognitive deficits do not interfere with functional independence but subtle difficulties on complex functional tasks may be present.	• Diagnosis of PD (according to Brain Bank criteria, Gibb & Lees, 1988) and dementia syndrome with insidious onset and slow progression in context of established PD. • Diagnosed by history, clinical, and mental examination, defined as: impairment in more than one cognitive domain; representing a decline from premorbid level; deficits severe enough to impair daily life (social, occupational, personal care), independent of the impairment ascribable to motor or autonomic symptoms.	• Dementia: progressive cognitive decline of sufficient to interfere with normal social, occupational or daily functioning. • Prominent or persistent memory impairment may not necessarily occur in the early stages but is usually evident with progression. • Deficits on tests of attention, executive function, and visuoperceptual ability may be especially prominent and occur early.
Associated clinical features	**Non-motor symptoms** *(Chaudhuri et al., 2011)* include: **Neuropsychiatric** *(some may be drug-induced)***:** apathy, depression, anxiety, cognitive impairment, hallucinations, delusions, impulse control disorder; **Sleep dysfunctions:** REM sleep behaviour disorder,	**Level I:** Impairment on a scale of global cognitive abilities. **Level II:** Neuropsychological testing that includes at least 2/5 of tests that measure attention and working memory, executive functioning, language, memory, or visuospatial abilities; *or* impairment on	**Probable:** ≥2 cognitive with or without behavioural. **Possible:** ≥ 1 atypical (e.g., prominent or receptive-type aphasia, pure amnesia with no benefit from cuing, preserved attention) cognitive with or without behavioural. **Cognitive:** impairment in attention, executive functions, memory,	**Probable:** ≥2 core clinical features with or without indicative biomarkers or 1 core feature with 1 or more indicative biomarkers. **Possible:** 1 core clinical feature without indicative biomarker or ≥ 1 indicative biomarkers with no core clinical features for possible. **Core clinical:** fluctuating cognition

	restless legs syndrome; **Sensory:** pain, olfactory or visual disturbances; **Gastrointestinal:** dribbling of saliva, dysphagia; **Other**: fatigue, bladder/sexual dysfunctions; fluctuations in cognitive/ psychiatric, sensory/pain, weight.	neuropsychological tests (if performance is 1-2 SDs below the norms or decline is seen on serial cognitive testing).	visuospatial functions, but language largely preserved. **Behavioural:** apathy, changes in personality and mood (i.e. depression, anxiety), excessive daytime sleepiness, hallucinations, delusions.	(with pronounced variations in attention and alertness), recurrent detailed and well-formed visual hallucinations, REM sleep behaviour disorder, spontaneous cardinal features of parkinsonism (i.e. bradykinesia, rest tremor or rigidity).
Supportive features	**≥3 required for diagnosing PD** • Unilateral onset • Rest tremor present • Progressive disorder • Persistent asymmetry affecting side of onset most • Excellent response (70-100%) to levodopa • Severe levodopa-induced chorea • Levodopa response for 5 years or more • Clinical course of 10 years or more		**Features** which make the diagnosis uncertain (if both absent, probable diagnosis of PDD, if 1 absent, possible): coexistence of other abnormality that by itself may cause cognitive impairment but judged not to be the cause of dementia, and/or time interval between motor and cognitive symptoms unknown. **Features** suggesting other conditions or diseases as cause of mental impairment, making it impossible to reliably diagnose PDD must be absent, such as acute confusion, major depression, features compatible with vascular dementia according to NINDS-AIREN criteria.	**Supportive clinical:** severe sensitivity to antipsychotic agents, postural instability, repeated falls, syncope or other transient episodes of unresponsiveness, severe autonomic dysfunction, hypersomnia, hyposmia, hallucinations in other modalities; systematized delusions, apathy, anxiety, and depression. **Supportive biomarkers:** relative preservation of medial temporal lobe structures on CT/MRI scan; generalized low uptake on SPECT/PET perfusion/ metabolism scan with reduced occipital activity 6 the cingulate island sign on FDG-PET imaging; prominent posterior slow-wave activity on EEG with periodic fluctuations in the pre-alpha/ theta range.

Abbreviations: CT scan – computed tomography; DLB – Dementia with Lewy bodies; EEG – electroencephalography; FDG-PET imaging – fluorodeoxyglucose positron emission tomography; MRI scan – magnetic resonance imaging; NINDS-AIREN – National Institute of Neurological Disorders and Stroke and Association Internationale pour la Recherche et l'Enseignement en Neurosciences; PD – Parkinson's disease; PD-MCI – Parkinson's disease and mild cognitive impairment; PDD – Parkinson's disease dementia; PET scan – positron emission tomography; REM – rapid eye movement sleep; SPECT scan – single-photon emission computed tomography.

Appendix C: Documents of the PhD studies

We welcome your help

Assessing iCST for people with movement disorders and their caregivers is extremely important to us. For example, we would like to know if the adapted therapy is acceptable and if it meets peoples' needs.

The INVEST project aims to gather this information by conducting an NIHR Research for Patient Benefit Programme funded study. If you would be interested in taking part and using the therapy at home please contact:

Dr Sheree McCormick: Tel: 0161 306 7494

sheree.mccormick@manchester.ac.uk

Ms Sabina Vatter: Tel: 0161 306 7913

sabina.vatter@manchester.ac.uk

If the adapted iCST is successful, it will improve the quality of life for people with movement disorders and their caregivers.

INVEST
IN PARKINSONIAN DEMENTIAS

Individual Cognitive Stimulation Therapy (iCST) for People with Movement Disorders

This information booklet is provided for:

- Individuals with Parkinson's disease dementia or Dementia with Lewy bodies
- Family members
- Caregivers

MANCHESTER 1824 The University of Manchester

NHS National Institute for Health Research

The Challenge

People with Parkinson's disease are 20 – 40% more likely to develop dementia compared to people without the disease. Dementia, or mild cognitive impairment, in Parkinson's disease can cause mental impairment, such as problems with memory and attention, and can have a negative impact on people with the disease and their caregivers.

Cognitive Stimulation Therapy has been demonstrated to help overcome some of the mental challenges that can decrease quality of life for people with dementia and their caregivers. The therapy involves engaging in structured, enjoyable activities that support memory and stimulate communication and everyday planning.

Cognitive Stimulation Therapy is the only non-drug therapy widely used in the NHS. However, it is unknown whether this therapy is suitable for people with Parkinson's disease dementia or Dementia with Lewy bodies.

Cognitive Stimulation Therapy can help overcome some of the mental challenges associated with dementia and improve quality of life

A Solution

The INVEST study has developed Individual Cognitive Stimulation therapy (iCST) for people with Parkinson's disease dementia or Dementia with Lewy bodies. iCST is delivered at home by the caregiver. The therapy involves structured activities that are interesting and enjoyable. The manual contains guides for each task and no additional equipment is required.

A typical therapy session might involve:

Selecting a topic of your choice and sharing your opinions about the photographs and images provided.

Recalling happy memories linked to the topic.

Using different senses to think about the topic.

If desired, completing more challenging activities related to the topic.

Do you have Parkinson's disease and dementia?

Do you care for someone with Parkinson's disease and dementia?

You might be eligible to take part in our study.

INVEST

IN PARKINSONIAN DEMENTIAS

Psychosocial Therapy to Benefit Patients with Parkinson's-related Dementia: A Feasibility and Exploratory Pilot Study (INVEST).

In recent years, Cognitive Stimulation Therapy (CST) groups have shown to be an enjoyable and beneficial therapy for people with memory problems. The idea is to keep the mind active through enjoyable activities.

The INVEST project will look at whether individualised (one-to-one) CST can be tailored for people who have problems with memory and also have difficulties with movement.

We are looking for people with Parkinson's disease with dementia or dementia with Lewy bodies and their caregivers to take part in a short trial of individualised CST. People with Parkinson's disease and "mild cognitive impairment" (less severe memory problems than dementia) may also be eligible.

Caregivers would need to be willing to give iCST sessions for 30 minutes, three times a week and would receive training to do so. The activities will include, for example, discussion of art and current affairs.

If you might be interested in more information about the INVEST study, please contact:

Dr Sheree McCormick on 0161 306 7492 or sheree.mccormick@manchester.ac.uk /

Ms Sabina Vatter on 0161 306 7913 or sabina.vatter@manchester.ac.uk

INVEST Advert Version 1.1 22.05.2017

INVEST

in Parkinsonian Dementias

Issue 1, Date: July, 2016

Contents

WELCOME

Welcome to the first edition of the INVEST newsletter. This quarterly newsletter is for the clinical, academic and research colleagues with whom we are proud to collaborate. Please do not share with participants or patients.

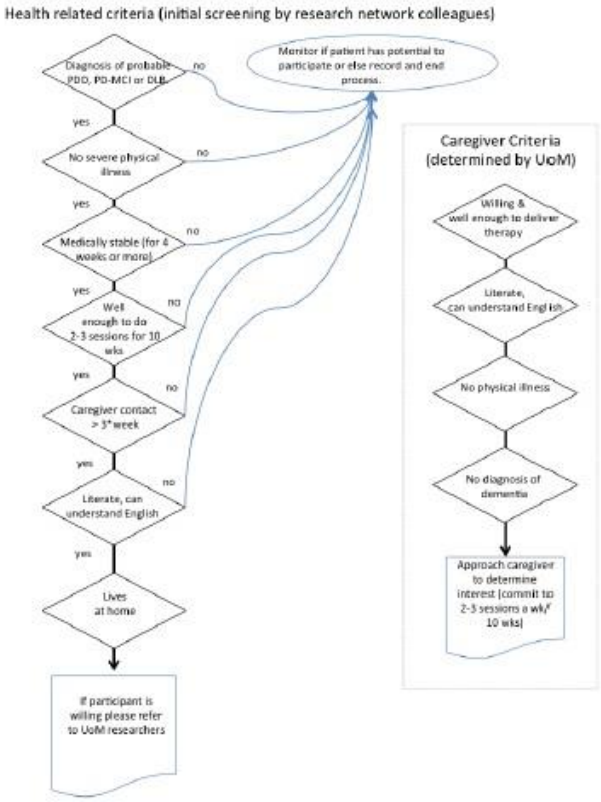
The INVEST trial is sponsored by Manchester Mental Health and Social Care Trust (MMHSCT), funded by the Research for Patient Benefit programme (competition 22) and supported by the Clinical Research Network. The trial is conducting vital preparatory work to support the efficacy testing of a psychosocial intervention specifically tailored for people living with Parkinsonian dementias and their caregivers.

We hope you enjoy the newsletter and thank you for your support.

Dr Sheree McCormick, Project Manager

Ms Sabina Vatter, Research Assistant

Recruitment Protocol



Recruitment protocol

- Research network colleagues (PIs, specialist PD nurses and DeNDRoN research nurses) identify potential participants and hand out/send out user-friendly information booklet
- Potential participants are encouraged take at least 24 hours to discuss participation with family and friends
- If interest is maintained, the Trial team:
  - Send out additional information (sample topics, participant information sheets)
  - Conduct home-based screening
- If eligibility is confirmed, informed consent is obtained and the dyad (participant with PDD, PD-MCI, or DLB and their caregiver) are assessed and randomised to either Cognitive Stimulation Therapy or Treatment as Usual.

Recruitment Update

We are recruiting participants from memory clinics across four Trusts: MMHSCT, Salford Royal NHS Foundation, Pennine Acute Hospitals NHS Trust (PAT) and University Hospital South Manchester (UHSM). Participants are also being identified through Join Dementia Research, Parkinson's UK and the Lewy Body Society

All four sites are now open to recruitment; MMHSCT opened on 10th February, Salford Royal opened on 8th March, PAT opened on 13th April and UHSM opened on 10th May.

Since recruitment to the main trial commenced (12th April, 2016) 12 dyads (24 participants) have been randomized; MMSCT have randomised 6, Salford Royal have randomized 2, PAT have randomized 1 and UHSM have randomized 3.

The bar chart on the right shows recruitment by centre and month since the start of the trial, the pie chart on the left shows distribution of randomization. Attrition rate (number withdrawn/number enrolled) is 15% (two participants have withdrawn).

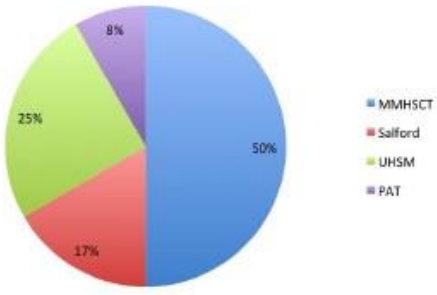
MMHSCT. PI is Dr Iracema Leroi.

Salford Royal NHS Foundation. PI is Dr Monty Silverdale.

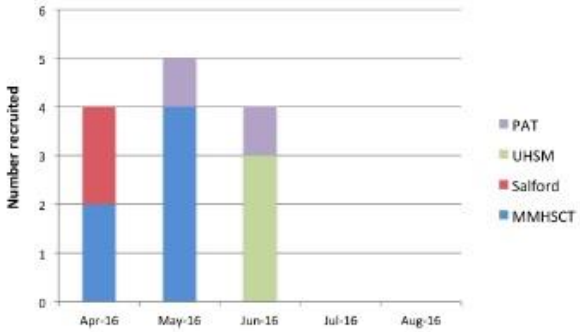
Pennine Acute Hospitals NHS Trust. PI is Dr Jason Raw.

University Hospital South Manchester. PI is Dr David Ahearn.

INVEST % of randomised participants as of 30th June 2016



INVEST Monthly Recruitment from 1st April 2016



Adapted Cognitive Stimulation Therapy

"I fear I am not in my perfect mind… What place this is; and all the skill I have remembers not these garments; nor I know not where I did lodge last night…" [King Lear by W. Shakespeare, Act IV, Scene 7]

Our concept for the INVEST study started over a decade ago. At that time, our team was undertaking clinical trials for memory enhancing medication to improve symptoms of dementia in Parkinson's disease (PD). While we saw some symptomatic improvements with the medications, we found that important aspects of people's lives were not being addressed by these types of treatments. In particular, support for the symptoms of apathy, changes in the relationship between the person with PD and their significant other, caregiver burden and overall quality of life remained untouched by any potential improvement in cognitive symptoms. More had to be done.

So, we looked around at different types of non-medication treatments, the so-called 'psychosocial therapies', to determine whether they might be applicable in PD. We analysed existing therapies for non-PD dementia and determined which ones might be adapted for PD. We chose 'cognitive stimulation therapy', (CST) an intervention that involves an affected person and their significant other in exciting dialogues and interactions. CST encourages and stimulates discussions and debates about a variety of topics. We secured funding for a pilot trial, consulted extensively with stakeholders about how the adaptation should look, and finally, in April 2016, enrolled our first study participant.

So far the pilot study has gone well and is on track to finish in 2017. We are already learning a lot about the feasibility and tolerability of the adapted therapy and hope to apply for funding for a subsequent definitive full scale trial. This study represents a significant step in bringing new opportunities for therapeutic interventions for people with dementia in PD.

Iracema Leroi MD FRCPC MRCPsych

Chief Investigator

The INVEST team are grateful to the contribution of Iván Sebastià (pictured above) who kindly provided his artwork for the therapy manual cover. Iván says: *"I have always liked art and crafts. I was diagnosed with Parkinson's over 10 years ago and after living with it for a few years I went to the Parkinson's Association where I discovered Art Therapy. I participated in an Art Therapy group for 6 years. It has helped me link art making with self-expression, opening up my mind and now I believe anything is possible. What motivates me to create is the curiosity of not knowing what the end result will be. If you focus on the process, art-making becomes an exciting journey and one that teaches you about life. I want to thank my wife and children for all their love and support."*

Time to **INVEST**

Jan 16

Ethics approved
Field testing completed
Therapy manual printed
MMHSCT & Salford approved

Apr 16

First participant randomised
Welcome visits to sites
INVEST presentation to MMHSCT
UHSM, PAT approved
PPI representatives join team

Jul 16

Oct 16

Jan 17 — Recruitment target = 50% (32 dyads)

Apr 17

Jul 17

Oct 17

November 2017 – recruitment completed

Thank you!

Many people have kindly supported the trial since ethical approval was granted in January 2016. The INVEST team would like to thank the following people who have contributed through participating research networks, trial units or NHS sites.

A BIG thank you to:

Heena Mistry, Lewis Harpin,

Ambily Mani, Anita Davies,

Emma Oughton, Phillip Tinkler,

Judith Brooke, Rebecca Davies,

Robert Bedford, Patsy Cotton,

Ailish Fountain, Carol Miller

for working closely with the INVEST team to actively promote the study and identify, recruit and assess participants.

An equally big thank you also goes to all of our Principal Investigators, members of our Trial Steering Committee, Wider Management Group, Data Monitoring and Ethics Committee, Research and Development colleagues. Finally we would like to acknowledge the commitment and support provided by Parkinson's UK, our Patient and Public Involvement representatives and most importantly all of our participants

Trial team contact details

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Sabina.Vatter@manchester.ac.uk

This newsletter highlights independent research funded by the National Institute for Health Research under its Research for Patient Benefit Programme (PB-PG-0613-31058). The views expressed are those of the authors and not necessarily those of the NHS, NIHR or the Department of Health.

INVEST

in Parkinsonian Dementias

Issue 2, Date: January 2017

Contents

WELCOME

Welcome to the second edition of the INVEST newsletter. This is an exciting time for the project. We are pleased to report that since our first issue we are now recruiting in two additional sites: North East London Foundation Trust (PI Joanne Rodda) and Derbyshire Foundation Healthcare Trust (PI Christine Taylor). In Manchester, we are delighted to welcome Ms Sophie Baker, research assistant, to the team.

Further good news is that we have been commended by our funders (NIHR Research for Patient Benefit Programme) for the diligent manner in which the study is being executed. Our website is also live (www.invest-trial.com) and provides an excellent portal for all study related information.

Despite these exciting developments, dyadic recruitment in PD remains as challenging as ever. **We need your help** if we are to meet our recruitment targets and deliver the project on time. The inclusion criteria for the trial can be found on page two, please do contact the team if you have any questions.

Thank you for your support

Dr Sheree McCormick, Project Manager

Inclusion Criteria

Participants with Parkinson's related dementia

Inclusion criteria

- Have a diagnosis of possible or probable MCI-PD, PDD or DLB. Diagnosis will be based on standard clinical diagnostic criteria determined by the referring clinician and verified on screening;
- Be willing to participate in 30 minute sessions of the intervention, three times per week;
- Be literate and have a good understanding of conversational English;
- Be on a stable medication regimen for at least four weeks, prior to study entry;
- Have sufficient physical and mental capability to participate in the therapy. Capability will be based on clinical impression and informed by scores obtained on the UPDRS, the H & Y scale and the MoCA.

Exclusion criteria

- Current involvement in any other dementia intervention research study;
- Living in residential care.

Study Companions

Inclusion criteria

- Provide care or support for the person with Parkinson's-related dementia;
- Be willing and well enough to deliver 30 minute sessions of the intervention, three times per week;
- Be able to provide feedback regarding the participant's physical and mental health;
- Be able to undertake the training and delivery of the intervention.

Exclusion criteria

- Diagnosis of dementia.

Recruitment Update

INVEST team members, Sheree McCormick, Lucy-Jane Castle and Sabina Vatter, celebrating the recruitment of their 50th participant (25th dyad), September 2016.

Our recruiting sites from January 2017 onwards are:

Greater Manchester Mental Health NHS Foundation Trust (previously Manchester Mental Health & Social Care Trust). Dr **Iracema Leroi** is the local PI (and CI of the study).

Salford Royal NHS Foundation Trust. The local PI is **Dr Monty Silverdale.**

The Pennine Acute Hospitals NHS Trust. The local PI is **Dr Jason Raw**.

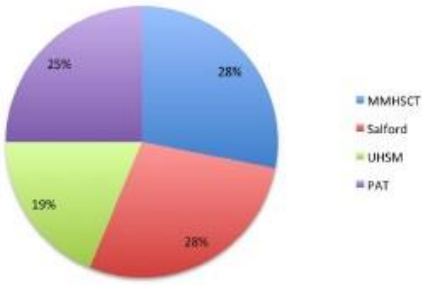
University Hospital of South Manchester NHS Foundation Trust. The local PI is **Dr David Ahearn.**

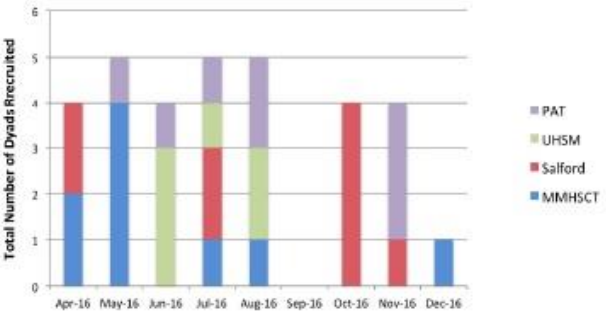
NEW-SITE: North East London Foundation Trust. The local PI is **Dr Joanne Rodda** with collaboration from **Ritchard Ledgerd.**

NEW-SITE: Derbyshire Healthcare Foundation Trust. The local PI is **Dr Christine Taylor.**

Since recruitment to the main trial commenced (12th April, 2016) 32 dyads (64 participants) have been randomised; MMHSCT have randomised 9, Salford Royal have randomised 9, PAT have randomised 8 and UHSM have randomised 6.

Percentage of recruited participants per site (Apr - Dec 2016)





Relationships in Parkinson's-related dementia

"Parkinson's has taken us on an unpredictable journey that changes daily. Sometimes it feels like we both have it. Despite this, our relationship is stronger because I care about him more."

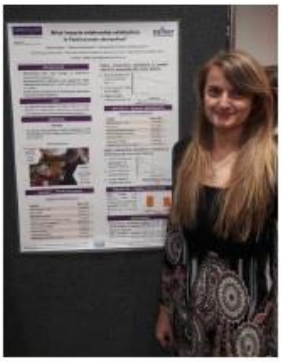
Loving relationships include feelings of closeness, connectedness, warmth and bondedness (Sternberg, 1997) as well as mutuality, reciprocity, commitment, intimacy, trust and communication. But what happens if one partner is diagnosed with a progressive neurodegenerative condition? How do their roles change? How does the relationship change?

Relationships in Parkinson's disease (PD) have been described as mutual and close but, due to the complex nature of the motor, psychiatric and cognitive symptoms, face challenges that can transform a partnership into 'care provider – care recipient' roles. This can lead to spouse's burden, stress and low mood which in turn can impact relationship satisfaction and quality.

Humans have an inherent motivation to form relationships with others. From time to time, however, relationships can become challenging whether those involved have a medical condition or not. A better understanding of if, and how, relationships change as cognitive impairment emerges in PD is essential to foster good outcomes in the condition.

Ms Sabina Vatter, research assistant on the INVEST project and 2nd year PhD student, is investigating the factors that impact on the marital relationship satisfaction in couples within Parkinson's-related cognitive impairment. Preliminary analyses from Sabina's research suggest people with PD report higher relationship satisfaction when there are fewer constraints on health. In spouses, lower levels of stress and burden are associated with greater relationship satisfaction.

These findings suggest relationship satisfaction needs to be considered when deigning interventions to support couples where one partner has PD.



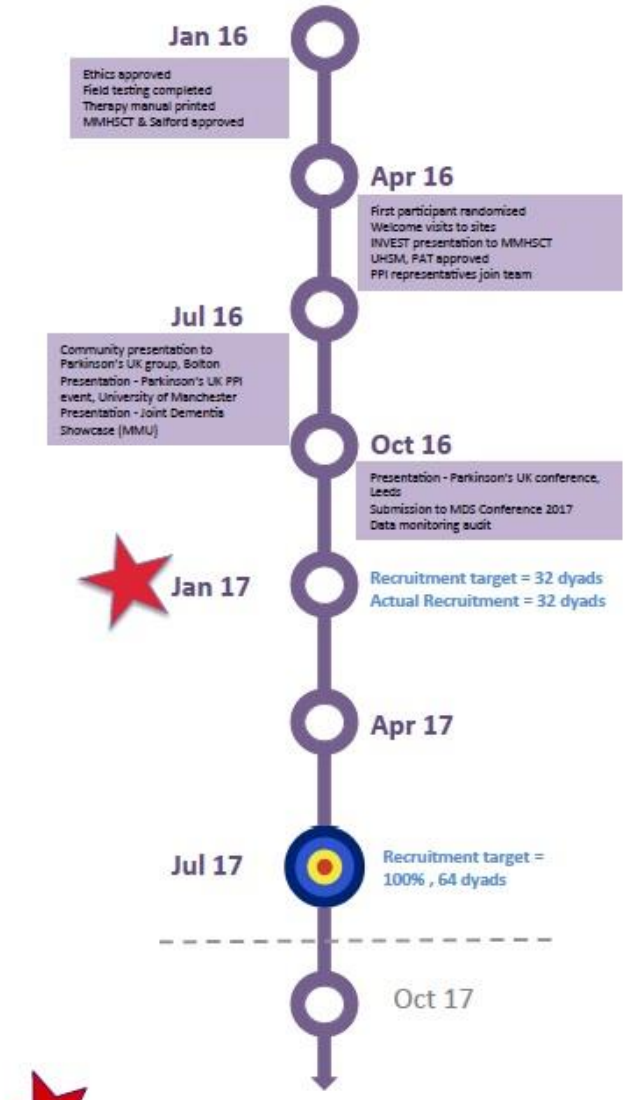
Sabina was recently awarded one of the coveted prizes at the launch of the School of Biological Sciences, University of Manchester, for her poster entitled "What impacts relationship satisfaction in Parkinsonian Dementias?"

Reflections on the therapy from a study companion

"I observed my husband and our grandson, it was very good to see them engaging and for our grandson to take the lead. They both enjoyed the session and the grandson said he had fun with Gramps"

A quote from one study companion highlighting the many benefits (e.g. self-efficacy, confidence, social interaction and relationship satisfaction) of the INVEST home-delivered psychosocial intervention for people with Parkinson's-related dementia.

Time to **INVEST**



It is anticipated that the addition of the two new sites (NELFT and Derbyshire) will allow the excellent recruitment rate demonstrated in 2016 to be maintained in 2017. The recruitment target (64 dyads) should be achieved as per the original protocol milestone (1st July 2017).

New Year, New Sites and New Staff

Please welcome our new researchers to the trial!

From left, Kate, Jane & Eanna, NELFT

From left, Jacki, Marta & Krisha, NELFT

Sophie Baker, University of Manchester

From left, Deborah, Lisa, Audrey, Diane & Caroline, Derbyshire Healthcare Foundation

Trial team contact details

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GMCRN.dementia@nihr.ac.uk quote INVEST

This newsletter highlights independent research funded by the National Institute for Health Research under its Research for Patient Benefit Programme (PB-PG-0613-31058). The views expressed are those of the authors and not necessarily those of the NHS, NIHR or the Department of Health.

The University of Manchester
Jean McFarlane Building
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Manchester
M13 9LP

..... of 2017

Dear .....,

Further to our recent conversation, please find enclosed a "Participant information sheet" and an "Information sheet for caregivers" about our movement disorder research at the University of Manchester. The aim of our research is to assess a non-drug based therapy called individual Cognitive Stimulation Therapy. The therapy aims to improve memory function and increase quality of life and has been specifically adapted for people with Parkinson's disease or Lewy Body dementia. The therapy is provided by the caregiver in a home-setting and is guided by a manual which contains a variety of discussion topics, activities and games. We have attached a sample topic to this letter.

As the therapy is delivered by the caregiver, it is important to consider if it is suitable for both of you. For example, would you both be able and willing to participate in 20-30 minutes of therapy, two to three times a week, for 10 weeks? We are happy to discuss this matter, and any other questions you might have, during our follow-up telephone call on .....

Prior to taking part in this research, you will be asked to give informed consent. Taking part in the study is voluntary and you are free to withdraw at any time.

If you have any questions please do not hesitate to contact either:

- Dr Sheree McCormick on 0161 306 7494 or sheree.mccormick@manchester.ac.uk or
- Ms Sabina Vatter on 0161 306 7913 or sabina.vatter@manchester.ac.uk

Thank you for considering taking part in our research, I look forward to talking to you soon.

Best wishes,

Sabina Vatter, on behalf of Dr Sheree McCormick and Dr. Iracema Leroi. (0161 306 7913)

Attached to the letter: Participant Information Sheet dated 18th of April 2017 (Version 1.4) & Information Sheet for Caregivers dated 18th of April 2017 (Version 1.4)

PARTICIPANT INFORMATION SHEET

Psychosocial Therapy to Benefit Patients with Parkinson's-related Dementia: Development of an individualised Cognitive Stimulation Therapy programme.

Invitation to participate in a research study

You are being invited to take part in a research study. Before you decide, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part. Thank you for reading this information sheet.

What is the purpose of the study?

In recent years, Cognitive Stimulation Therapy (CST) groups have shown to be an enjoyable and beneficial therapy for people with memory problems. This project will show whether individualised (one-to-one) CST can be tailored for people who have problems with memory and also have difficulties with movement (for example, a tremor or muscle stiffness).

Why have I been chosen?

You have been invited to take part because you have had a diagnosis of a movement disorder with memory problems. We need up to seventy-six people in total with a movement disorder and memory problems for this study.

What happens in individualised cognitive stimulation therapy (iCST)?

iCST sessions will last for 30 minutes and will be led by your relative/friend. They will take place three times a week for ten weeks. The activities will include, for example, discussion of art and current affairs. The idea is to keep the mind active through enjoyable activities.

Do I have to take part?

It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time without giving a reason. A decision to withdraw at any time, or a decision not to take part, will not affect the standard of care you receive.

What happens to me if I take part in this study?

This study is a randomised trial. We need to see whether iCST is better than no treatment, so we need to compare any changes experienced by people receiving iCST to those receiving no additional therapy. The fairest way of doing this is to select people for the group by chance; everyone agreeing to take part will have a 50:50 chance of receiving iCST. The decision is made by an independent computer, which will not have any identifying information about you or your relative/friend.

If you decide to take part the study will last for a time period of about fourteen weeks. Following discussion of any questions you may have with a researcher, and signing the consent form, all participants will be allocated to the iCST group or the group that doesn't receive iCST, then:

1. All participants will be asked to meet with a researcher for between two / two-and-a-half hours for an interview and to complete some questionnaires covering your quality of life, cognition (e.g. memory) and mood. The time stated to complete the interviews and assessments is an estimate; you and your friend/relative may take as many breaks as you want or feel necessary, and even complete the process over two sessions if preferred.

2. *For the iCST group:* complete three sessions of iCST, each lasting 30 minutes, per week, for ten weeks, *or for the group that doesn't receive iCST:* continue your regular daily activities without the therapy sessions.

3. All participants will be asked to repeat the questionnaires and assessments with the researcher after 10 weeks. This is to see whether any of these factors changed for people who received the iCST intervention. At this point (if you received iCST) you may also be asked to complete a separate interview with the researcher to record what you thought about the therapy. It is important for you to know that this interview will be voice recorded for analysis at a later date.

Usually, the researcher will come to your home or the home of your relative/friend, but will be happy to meet you elsewhere if you would prefer. The researcher will meet with and interview your relative/friend at the same time as you are completing the questionnaires.

Optional parts of the study:

1. After completing the study, participants allocated to the control group will be offered an opportunity to experience the therapy. A researcher will visit the participants at home and provide therapy training. Following therapy training the participants will have access to on-going support from the research team for a period of 10 weeks. No data will be collected during or after this time, the therapy is offered as a courtesy to the control group.

Optional parts of the study (continued):

2. At the end of the study, we may ask some participants to take part in focus groups. These will last around half a day and will be led by the research team. The purpose of the focus groups is to ask for your feedback on how the therapy might be improved or adapted. Refreshments will be provided for everyone during breaks. It is important for you to know that the focus group will be voice recorded for analysis at a later date.

Expenses
Any reasonable travel expenses incurred by you or your care-giver will be reimbursed.

What do I have to do?
Taking part in the study does not involve any lifestyle restrictions or changes. You can carry on your everyday activities as normal. All we ask is that you keep your appointments with us on the days of the assessments and the focus group.

What are the possible disadvantages and risks of taking part?
iCST aims to be stimulating and enjoyable. Sessions involve discussing themes such as food, childhood and current affairs and the level of risk in taking part is therefore minimal. Your caregiver will be given guidance on what to do if you become anxious or distressed during sessions. If the intervention really does not suit you, you are free to finish or withdraw from the study at any point.

What are the possible benefits of taking part?
If you decide to take part we hope that it might be enjoyable for you. There may be no direct benefit to participating but we hope that the stimulating activities might improve your quality of life. The information that we get from this study may help us to develop a programme that, in the future, could help to treat people with movement disorders and memory problems. As such, your contribution would be valuable.

Will my taking part in the study be kept confidential?
All information which is collected about you during the course of the study will be kept strictly confidential. All data is stored without any identifying details under secure conditions.

Will my GP be informed of my involvement?
Yes, we will ask for your permission to send your GP a letter explaining that you have agreed to take part in the study.

What will happen if I don't want to carry on with the study?
You will be free to withdraw from the study at any time, without giving a reason. Withdrawing from the study will not affect the standard of care you receive. We will need to use any data collected in the study, up to the point of withdrawal.

What if something goes wrong?
If you are harmed by taking part in this study, there are no special compensation arrangements. If you are harmed due to someone's negligence, then you may have grounds for a legal action, but you may have to pay for your legal costs.

Regardless of this, if you wish to make a complaint about any aspect of the way you have been approached or treated during the course of this study, the normal National Health Service complaints procedures should be available to you. If you are unhappy or dissatisfied about any aspect of your participation, we would ask you to tell us about this in the first instance, so that we can try to resolve any concerns and find a solution.

Who is organising and funding the research?
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Consent form for use of audio recording and direct quotes
An audio recording of interviews and focus groups will be taken. The purpose of the audio recording is to help the researchers to identify the most important aspects of the discussion at a later date. You may at any point request that the audio recording is paused, or temporarily stopped. However, we will require use of the recording to this point. We may use direct quotes from the audio recording in our publications but we will do so in an anonymised form so that your identity is protected.

What will happen to the results of the research?
The results from this study will help us decide whether or not to run a larger study to help people with movement disorders and cognitive impairment. The results will be published by the Department of Health, and in relevant health journals. No participants will be identified in any publication arising from the study, without their written consent. We will make arrangements for participants to be informed of the progress of the research and the results through newsletters and local meetings. If you would like a short summary at the end of the study please let the research team know. Participants in the control arm will also be able to access the research findings and receive a short summary of the results if they wish. Upon completion, a summary can be posted at your home address.

Who has reviewed the study?
All NHS research is looked at by an independent group of people, called a Research Ethics Committee to protect your safety, rights, well-being and dignity. This study has been reviewed and been given a favourable opinion by the NRES Yorkshire & The Humber – Bradford Leeds Research Ethics Committee.

Who can I contact for further information?
If you would like further information or have any questions about the study please contact one of the study researchers listed below:

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Ms Sabina Vatter
Research Assistant
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Or if you have any complaints about this study please contact:
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If there are any issues regarding this research that you would prefer not to discuss with the research team, please contact the Research Practice and Governance Co-ordinator by either writing to 'The Research Practice and Governance Co-ordinator, Research Office, Christie Building, The University of Manchester, Oxford Road, Manchester, M13 9PL', by emailing research.complaints@manchester.ac.uk or by telephoning 0161 275 8093.

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Thank you for considering taking part in this research study.

INFORMATION SHEET FOR CAREGIVERS

Psychosocial Therapy to Benefit Patients with Parkinson's-related Dementia: Development of an individualised Cognitive Stimulation Therapy programme.

Invitation to participate in a research study
You are being invited to take part in a research study. Before you decide, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part. Thank you for reading this information sheet.

What is the purpose of the study?
In recent years, Cognitive Stimulation Therapy (CST) groups have shown to be an enjoyable and beneficial therapy for people with memory problems. This project will show whether individualised (one-to-one) CST can be tailored for people who have problems with memory and also have difficulties with movement (for example, a tremor or muscle stiffness).

Why have I been chosen?
You have been invited to take part because of your support for a person who has had a diagnosis of a movement disorder with memory problems. We need up to seventy-six people in total who are caregivers for people with movement disorders and memory problems.

What happens in individualised cognitive stimulation therapy (iCST)?
iCST sessions will last for 30 minutes and take place three times a week for ten weeks. The activities will include, for example, discussion of art and current affairs. The idea is to keep the mind active through enjoyable activities. We will ask you to guide your relative/friend through the activities and will provide you with the training and support you need to be able to do so. We will ask you to complete provide some about each of the activities.

Do I have to take part?
It is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time without giving a reason. A decision to withdraw at any time, or a decision not to take part, will not affect the standard of care your relative / friend receives.

What happens to me if I take part in this study?

This study is a randomised trial. We need to see whether iCST is better than no treatment, so we need to compare any changes experienced by people receiving iCST to those receiving no additional therapy. The fairest way of doing this is to select people for the group by chance; everyone agreeing to take part will have a 50:50 chance of receiving iCST. The decision is made by an independent computer, which will not have any identifying information about you or your relative/friend.

If you decide to take part, stage one of the study will last for a time period of about fourteen weeks. Following discussion of any questions you may have with a researcher, and signing the consent form, all participants will be allocated to the iCST group or the group that doesn't receive iCST, then:

1. If you are in the iCST group you will be asked to meet with a researcher for a half day of training to lead the iCST sessions. The training can take place in your home or at the University of Manchester, if you prefer. This will happen around two weeks before starting the therapy, to give you time to think, read and ask any questions you might have.

2. All participants will be asked to meet with a researcher for between two / two-and-a-half hours to complete an interview and some questionnaires covering your quality of life, mood, and about the relationship you have with your relative/friend. You will also be asked to complete some health questions about your relative/friend. The time stated to complete the interviews and assessments is an estimate; you and your friend/relative may take as many breaks as you want or feel necessary, and even complete the process over two sessions if preferred.

3. *For the iCST group:* complete three sessions of iCST, each lasting 30 minutes, per week, for ten weeks, *or for the group that doesn't receive iCST:* continue your regular daily activities with your relative/friend without the therapy sessions.

4. All participants will be asked to repeat the questionnaires and assessments with the researcher after 10 weeks. This is to see whether any of these factors changed for people who received the iCST intervention. At this point (if your relative/friend received iCST) you may also be asked to complete a separate interview with the researcher to record what you thought about the therapy. It is important for you to know that this interview will be voice recorded for analysis at a later date.

Usually, the researcher will come to your home or the home of your relative/friend, but will be happy to meet you elsewhere if you would prefer. The researcher will meet with and interview your relative/friend at the same time as you are completing the questionnaires.

Optional parts of the study:

1. After completing the study, participants allocated to the control group will be offered an opportunity to experience the therapy. A researcher will visit the participants at home and provide therapy training. Following therapy training the participants will have access to on-going support from the research team for a period of 10 weeks. No data will be collected during or after this time, the therapy is offered as a courtesy to the control group.

2. At the end of the study, we may ask some participants to take part in focus groups. These will last around half a day and will be led by the research team. The purpose of the focus groups is to ask for your feedback on how the therapy might be improved or adapted. Refreshments will be provided for everyone during breaks. It is important for you to know that the focus group will be voice recorded for analysis at a later date.

Expenses
Any reasonable travel expenses you incur will be reimbursed.

What do I have to do?
Taking part in the study does not involve any lifestyle restrictions or changes. You can carry on with your everyday activities as normal. All we ask is that you keep your appointments with us on the days of the assessments, interviews and the focus group.

What are the possible disadvantages and risks of taking part?
iCST aims to be stimulating and enjoyable. Sessions involve discussing themes such as food, childhood and current affairs and the level of risk in taking part is therefore minimal. You will be given guidance on what to do if your relative/friend becomes anxious or distressed during sessions. If delivering the intervention really does not suit you or the intervention does not suit your friend/relative, you are free to finish or withdraw at any point.

What are the possible benefits of taking part?
If you decide to take part we hope that it might be enjoyable for you. There may be no direct benefit to participating but we hope that the stimulating activities might improve your quality of life. The information that we get from this study may help us to develop a programme that, in the future, could help to treat people with movement disorders and memory problems. As such, your contribution would be valuable.

Will my taking part in the study be kept confidential?
All information which is collected about you during the course of the study will be kept strictly confidential. All data is stored without any identifying details under secure conditions.

What will happen if I don't want to carry on with the study?
You will be free to withdraw from the study at any time, without giving a reason. Withdrawing from the study will not affect the standard of care you receive. We will need to use any data collected in the study, up to the point of withdrawal.

What if something goes wrong?
If you are harmed by taking part in this study, there are no special compensation arrangements. If you are harmed due to someone's negligence, then you may have grounds for a legal action, but you may have to pay for your legal costs.

Regardless of this, if you wish to make a complaint about any aspect of the way you have been approached or treated during the course of this study, the normal National Health Service complaints procedures should be available to you. If you are unhappy or dissatisfied about any aspect of your participation, we would ask you to tell us about this in the first instance, so that we can try to resolve any concerns and find a solution.

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What will happen to the results of the research?
The results from this study will help us decide whether or not to run a larger study to help people with movement disorders and cognitive impairment. The results will be published by the Department of Health, and in relevant health journals. No participants will be identified in any publication arising from the study, without their written consent. We will make arrangements for participants to be informed of the progress of the research and the results through newsletters and local meetings. If you would like a short summary at the end of the study please let the research team know. Participants in the control arm will also be able to access the research findings and receive a short summary of the results if they wish. Upon completion, a summary can be posted at your home address.

Who has reviewed the study?
All NHS research is looked at by an independent group of people, called a Research Ethics Committee to protect your safety, rights, well-being and dignity. This study has been reviewed and been given a favourable opinion by the NRES Yorkshire & The Humber-Bradford Leeds Ethics Committee.

Who can I contact for further information?
If you would like further information or have any questions about the study please contact one of the study researchers listed below:

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Research Associate
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sheree.mccormick@manchester.ac.uk

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Thank you for considering taking part in this research study.

Psychosocial Therapy to Benefit Patients with Parkinson's-related Dementia: Development of an individualised Cognitive Stimulation Therapy programme.

Information for Personal Consultee

Introduction

Your relative/friend has been invited to take part in a research study and we wish to seek your advice. We feel that your relative/friend's current illness prevents him/her from making an informed decision about taking part.

We would like you to advise us whether you think your relative/friend would be happy to take part in the research. Before advising us, please read this information sheet so that you know what is involved and feel free to ask questions. The information below is the same as would have been provided to your relative/friend. Thank you for reading this information sheet.

What is the purpose of the study?

In recent years, Cognitive Stimulation Therapy (CST) groups have shown to be an enjoyable and beneficial therapy for people with memory problems. This project will show whether individualised (one-to-one) CST can be tailored for people who have problems with memory and also have difficulties with movement (for example, a tremor or muscle stiffness).

iCST sessions will last for 30 minutes and will be led by someone close to your relative/friend. They will take place three times a week for ten weeks. The activities will include, for example, discussion of art and current affairs. The idea is to keep the mind active through enjoyable activities.

Why are you interested in clinical information about my relative/friend?

Your relative/friend has had a diagnosis of a movement disorder with memory problems. We need seventy-six people in total for this study.

Does my relative/friend have to take part?

It is up to you to decide whether or not your relative/friend would like to take part. If you do decide they would like to participate, you will be given this information sheet to keep and we will ask you to read and sign the Personal Consultee declaration form. You are still free to withdraw your relative/friend at any time without giving a reason. A decision to withdraw at any time, or a decision not to take part, will not affect the standard of care your relative/friend receives.

What will happen if I advise that my relative/friend would want to take part in this study?

This study is a randomised trial. We need to see whether iCST is better than no treatment, so we need to compare any changes experienced by people receiving iCST to those receiving no additional therapy. The fairest way of doing this is to select people for the group by chance; everyone agreeing to take part will have a 50:50 chance of receiving iCST. The decision is made by an independent computer, which will not have any identifying information about you or your relative/friend.

If you decide that your relative/friend would want to take part, stage one of the study will last for a time period of about fourteen weeks. Following discussion of any questions with a researcher, and signing the declaration form, all participants will be allocated to the iCST group or the group that doesn't receive iCST, then:

1. All participants will be asked to meet with a researcher for between two / two-and-a-half hours for an interview and to complete some questionnaires covering quality of life, cognition (e.g. memory) and mood. The time stated to complete the interviews and assessments is an estimate; your friend/relative may take as many breaks as they want or feel necessary, and even complete the process over two sessions if preferred.

2. *For the iCST group:* complete three sessions of iCST, each lasting 30 minutes, per week, for ten weeks, *or for the group that doesn't receive iCST:* continue their regular daily activities without the therapy sessions.

3. All participants will be asked to repeat the questionnaires and assessments with the researcher after 10 weeks. This is to see whether any of these factors change as a result of the iCST intervention. At this point (if your relative/friend received iCST) your relative/friend may also be asked to complete a separate interview with the researcher to record what they thought about the therapy. It is important for you to know that this interview will be voice recorded for analysis at a later date.

Usually, the researcher will come to your home or the home of your relative/friend, but will be happy to meet you elsewhere if you would prefer.

Optional parts of the study

1. After completing the study, participants allocated to the control group will be offered an opportunity to experience the therapy. A researcher will visit the participants at home and provide therapy training. Following therapy training the participants will have access to on-going support from the research team for a period of 10 weeks. No data will be collected during or after this time, the therapy is offered as a courtesy to the control group.

2. At the end of the study, we may ask some participants to take part in focus groups. These will last around half a day and will be led by the research team. The purpose of the focus groups is to ask for feedback on how the therapy might be improved or adapted. Refreshments will be provided for everyone during breaks. It is important for you to know that the focus group will be voice recorded for analysis at a later date.

Expenses
Any travel expenses incurred by your relative/friend or their caregiver will be reimbursed.

What does my relative/friend have to do?
Taking part in the study does not involve any lifestyle restrictions or changes. Your relative/friend can carry on their everyday activities as normal. All we ask is that your relative/friend keeps the appointments with us on the days of the interviews, assessments and focus groups.

What are the possible disadvantages and risks of taking part?
iCST aims to be stimulating and enjoyable. Sessions involve discussing themes such as food, childhood and current affairs and the level of risk in taking part is therefore minimal. The person delivering the therapy will be given guidance on what to do if your relative/friend becomes anxious or distressed during sessions. If the intervention really does not suit your friend/relative, they are free to finish or withdraw from the study at any point.

What are the possible benefits of taking part?
If you decide your relative/friend would like to take part we hope that it might be enjoyable for them. There may be no direct benefit to participating but we hope that the stimulating activities might improve your friend/relative's quality of life. The information that we get from this study may help us to develop a programme that, in the future, could help to treat people with movement disorders and memory problems. As such, your relative/friend's contribution would be valuable.

What will happen if your relative/friend doesn't want to carry on with the study?
Your relative/friend will be free to withdraw from the study at any time, without giving a reason. Withdrawing from the study will not affect the standard of care your relative/friend receives. We will need to use any data collected in the study, up to the point of withdrawal.

Will my relative/friend taking part in the study be kept confidential?
We will ask for your agreement to send your relative/friend's GP a letter explaining that they are taking part in the study. All information which is collected about your relative/friend during the course of the study will be kept strictly confidential. All data is stored without any identifying details under secure conditions.

Will my advice affect my relative/friend's clinical care?
No. If you decide that your relative/friend would not wish to participate in this study, this will in no way affect his/her clinical care.

Can I change my advice?
Yes, certainly. If at any time you feel that your advice was not in accordance with your relative/friends wishes please let us know.

What if something goes wrong?
If your relative/friend is harmed by taking part in this study, there are no special compensation arrangements. If your relative/friend is harmed due to someone's negligence, then your relative/friend may have grounds for a legal action, but your relative/friend may have to pay for their legal costs.

Regardless of this, if you or your relative/friend wish to make a complaint about any aspect of the way you have been approached or treated during the course of this study, the normal National Health Service complaints procedures should be available to you. If you or your relative/friend are unhappy or dissatisfied about any aspect of your participation, we would ask you to tell us about this in the first instance, so that we can try to resolve any concerns and find a solution.

Who is organising and funding the research?
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What will happen to the results of the research?
The results from this study will help us decide whether or not to run a larger study to help people with movement disorders and cognitive impairment. The results will be published by the Department of Health, and in relevant health journals. No participants will be identified in any publication arising from the study, without their written consent. We will make arrangements for participants to be informed of the progress of the research and the results through newsletters and local meetings. If you would like a short summary at the end of the study please let the research team know. Participants in the control arm will also be able to access the research findings and receive a short summary of the results if they wish. Upon completion, a summary can be posted at your home address.

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CONFIDENTIAL

Project Title:
Psychosocial Therapy to Benefit Patients with Parkinson's-related Dementia: Development of an individualised Cognitive Stimulation Therapy programme.

Chief Investigator: Dr. Iracema Leroi
Participant ID:

PLEASE INITIAL BOX

1. I confirm that I have read and understood the Participant Information Sheet dated 18th of April 2017 (version 1.4) for the above study and have had the opportunity to ask questions. ☐
2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my medical care or legal rights being affected. ☐
3. I agree to take part in the above study. ☐
4. If allocated to the treatment group, I agree to complete the therapy sessions three times per week for ten weeks. ☐
5. I agree to take part in the interviews and understand they will be audio recorded. ☐
6. I agree to the use of anonymised quotes in publications. ☐
7. I agree to my GP being informed of my participation in the study. ☐
8. I understand that if my capacity to make my own decisions changes during the study period, members of the research team may seek advice from a Consultee about using data collected from me for the purposes of the study and to ask about my continuation in participating. ☐

9. a) I agree to be contacted regarding participation in the focus group that will take place near the end of the study. ☐
(Optional) Yes or No: .....

b) I agree to the focus group consultation being audio recorded. ☐

c) I understand that if I withdraw during or after the focus group, the audio recording will still be used as it will contain information provided by other people. ☐

10. I agree to be contacted about future studies: ☐
(Optional) Yes or No: .....

Name of participant	Date	Signature
Name of researcher	Date	Signature

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PLEASE INITIAL BOX

1. I confirm that I have read and understood the Participant Information Sheet dated 18th of April 2017 (version 1.4) for the above study and have had the opportunity to ask questions. ☐
2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my medical care or legal rights being affected. ☐
3. I agree to take part in the above study. ☐
4. I agree to complete the caregiver training and, if allocated to the treatment group, delivering the therapy sessions three times per week for ten weeks. ☐
5. I agree to take part in the interviews and understand they will be audio recorded. ☐
6. I agree to the use of anonymised quotes in publications. ☐
7. a) I agree to be contacted regarding participation in the focus group that will take place near the end of the study. ☐
   (Optional) Yes or No: .....
   b) I agree to the focus group consultation being audio recorded. ☐

7. c) I understand that if I withdraw during or after the focus group, the audio recording will still be used as it will contain information provided by other people. ☐

8. I agree to be contacted about future studies: ☐

(Optional) Yes or No: .....

□

Name of participant	Date	Signature
Name of researcher	Date	Signature

CONFIDENTIAL

Psychosocial Therapy to Benefit Patients with Parkinson's-related Dementia: Development of an individualised Cognitive Stimulation Therapy programme.

Personal Consultee Declaration Form

I, _____, have been consulted about whether _____ would like to take part in the above study.

I have read the Personal Consultee information sheet dated 18th of April 2017 (version 1.4) and have had the opportunity to ask questions.

Please initial the box to confirm

1. In my opinion he/she would be happy to take part.
I understand that I can request that he/she is withdrawn at any time, without giving any reason, without his/her medical care or legal rights being affected. ☐

2. In my opinion he/she would agree to complete three therapy sessions per week, if allocated to the treatment group. ☐

3. In my opinion he/she would agree to complete the interviews.
I understand that the interviews are recorded. ☐

4. In my opinion he/she would agree to the use of anonymised quotes in publications. ☐

5. In my opinion he/she would agree to be contacted regarding participation in the focus group and would agree to the consultation being audio recorded.
I understand that if he/she is withdrawn during or after the focus group, the audio recording will still be used as it will contain information provided by other people. ☐

6. In my opinion he/she would agree to be contacted about future studies: *(Optional) Yes or No:* ..... ☐

_____	_____	_____
Name of Consultee	Date	Signature

Personal relationship of Consultee to Participant _____

_____	_____	_____
Name of researcher	Date	Signature

CONFIDENTIAL

Psychosocial Therapy to Benefit Patients with Parkinson's-related Dementia: Development of an individualised Cognitive Stimulation Therapy programme.

Consultee Declaration for Continued Participation Form

I, _____, have been consulted about whether _____ would like to remain in the above study.

I have read the Personal Consultee information sheet dated 18th of April 2017 (version 1.4) and have had the opportunity to ask questions.

Please initial the box to confirm

- I agree to _____'s continued participation in the above study, and for data collected since loss of capacity to be used for the purposes of the study. ☐

I understand that I can request that he/she is withdrawn at any time, without giving any reason, without his/her medical care or legal rights being affected.

_____ _____ _____

Name of Consultee Date Signature

Relationship (Professional or Personal) of Consultee to Participant:

_____

Please initial the box to confirm

- I have explained the study to the consultee and why they have been approached to provide agreement for _____ to remain involved in the study. ☐

_____ _____ _____

Name of researcher Date Signature

GP LETTER

Dr. Iracema Leroi
3rd Floor, Jean McFarlane Building
University of Manchester
Oxford Road
Manchester
M13 9PL
Iracema.leroi@manchester.ac.uk

..... of 2017

GENERAL PRACTITIONER INFORMATION SHEET

Title: Individualised Cognitive Stimulation Therapy (iCST) study

..... (DOB) ..... has been invited and consented to take part in a research study. Please let us know if there is anything that is not clear, or if you would like more information.

Dr. Iracema Leroi runs this project from the Manchester Mental Health and Social Care Trust and the University of Manchester.

Cognitive Stimulation Therapy (CST) groups are an enjoyable and beneficial therapy for people with dementia, recommended by the NICE (2007) guidelines. They aim to keep the mind active through enjoyable activities, which are undertaken as a structured programme facilitated by experienced and trained staff. However, many people do not have access to, or are not suited to group treatment.

This study will evaluate the impact of caregiver-led, individualised CST (iCST) on cognition and quality of life for people with movement disorders and dementia or cognitive impairment. It will involve three weekly sessions for ten weeks, covering similar themes to group CST (for example discussion of current affairs, food, word games). Caregivers will receive training and ongoing support in order to deliver the intervention effectively. It is a randomised controlled trial, therefore half the people participating will be allocated to a 'no treatment' control group, and will just be required to complete the assessment interview. After completion of the ten week period participants will have the option of completing a further ten weeks (so those who were not in the original treatment group have the possibility of receiving the therapy).

The assessments will be conducted prior to the intervention and after 10 weeks. There will be a 20 week assessment for those continuing in the trial. They will include outcome measures looking at:

- Personal details (age, relationship, medication, educational level, etc.)
- Quality-of-life (for both the person and their caregiver)
- Cognition
- Apathy
- Depression
- Activities of daily living and behaviour
- Caregiver mental health

The study will **not** affect your patient's current or future treatment.

The results of this study are expected to be published in relevant journals and at conferences. All interviews are confidential and will not be disclosed to anyone else. The information collected in the study will be anonymous and patients will not be identified in any report/publication. We may use direct quotes but these will also be confidential.

All proposals for research using human subjects are reviewed by the local Ethics Committee before they can proceed and the appropriate permission.

Thank you for reading this information sheet. Please do not hesitate to contact Dr. Leroi if you need any further information.

Kind regards,

_____

Research Assistant

Division of Neuroscience and Experimental Psychology
Faculty of Biology, Medicine and Health
3rd Floor Jean McFarlane Building
Oxford Road
Manchester
M13 9PL

DATE

Dear .....,

INVEST Study - Individual Cognitive Stimulation Therapy (iCST)

Thank you for agreeing to take part in the INVEST study. As you are aware you have been allocated to the iCST group. This means that both of you will be participating in the Individual Cognitive Stimulation Therapy (iCST) sessions.

iCST involves engaging in activities such as discussion topics and word games with the person you are caring for. We have enclosed a document outlining the nine principles that the therapy is based on. You may wish to read through this information prior to the training visit, which will take place on the _____. During the training visit you will receive all the resources you require for the therapy sessions and you will be receiving continuous support via weekly phone calls (and visits, should you require them) throughout the programme.

The Dendron research team will visit you again in 10 weeks (after the programme is completed) to repeat the questionnaire and assessment process. Please remember that **it is important that you do not tell them that you have been using iCST therapy**.

If you need to discuss any aspect of the study, please contact:

- Dr Sheree McCormick on 0161 306 7494 or sheree.mccormick@manchester.ac.uk or
- Ms Sabina Vatter on 0161 306 7913 or sabina.vatter@manchester.ac.uk

Thank you again for taking part in this research study, your help is greatly appreciated.

Yours sincerely,

Sabina Vatter

Sheree McCormick and Sabina Vatter
Jean McFarlane Building
University of Manchester
Oxford Road
Manchester
M13 9LP

DATE

Dear Mr & Mrs ……,

Individual Cognitive Stimulation Therapy (iCST) study,

Thank you for agreeing to take part in the iCST study. As you are aware you have been allocated to the control group. This means that both you and the person you are caring for will be continuing with your usual activities and not participating in the Individual Cognitive Stimulation Therapy (iCST) sessions.

However, you are an important part of the research process, so we still require you to complete the questionnaires and assessments in 10 weeks which will be arranged by the Dendron research team. Please remember that **it is important that you do not tell them that you are not participating in iCST.** The reason we ask the same questions again is to compare the results with the group that is receiving Individual Cognitive Stimulation Therapy (iCST).

At the ten-week point we will contact you to offer you the opportunity to undertake the Individual Cognitive Stimulation Therapy (iCST) sessions.

If you need to discuss any aspect of the study, please contact

- Dr Sheree McCormick on 0161 306 7494 or sheree.mccormick@manchester.ac.uk or
- Ms Sabina Vatter on 0161 306 7913 or sabina.vatter@manchester.ac.uk

Thank you again for taking part in this research study, your help is greatly appreciated.

Yours sincerely,

Sheree McCormick and Sabina Vatter

Reporting Serious Adverse Events in iCST

Instructions

1. Upon becoming aware of an adverse event involving a participant or caregiver, determine whether it is "serious" by examining the criteria below.

   A Serious Adverse Event (SAE) is an untoward occurrence experienced by either a participant or carer which:

   a) results in death;
   b) is life-threatening;
   c) requires hospitalisation or prolongation of existing hospitalisation;
   d) results in persistent or significant disability or incapacity;
   e) is otherwise considered medically significant by the investigator.

2. If a Serious Adverse Event is deemed to have taken place, please complete the attached form and forward it to the Chief Investigator (CI), Dr. Iracema Leroi, as instructed therein.

   It should be noted that all Serious Adverse Events should be reported to the CI, even if initially there may be no obvious connection to the trial. In particular:

   All deaths of participants and caregivers should be reported to the CI.

   All incidents of hospitalisation (and prolongation of hospitalisation) for participants and carers should be reported to the CI (even when the illness or condition being treated has no connection to the trial).

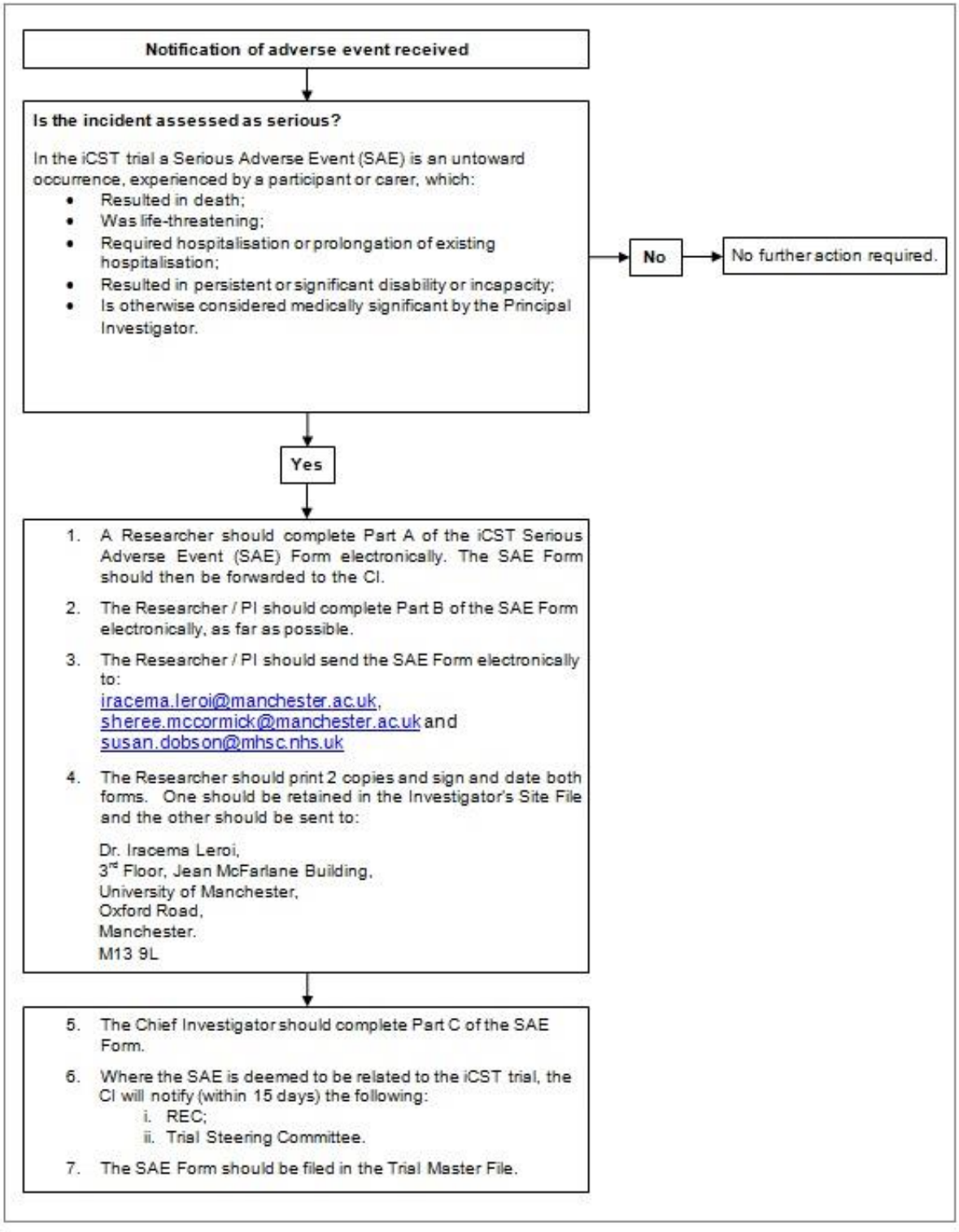
3. The INVEST Trial Steering Committee has specifically requested that, as far as possible, all hospitalisations are recorded. Researchers undertaking follow-up assessments (this does not apply to baseline assessments) should, therefore, consider this when completing the questionnaire booklet (Service Use). A SAE form should be completed where the participant or carer has indicated that they have stayed in hospital and this has not already been reported to the research team.

Has there been any Serious Adverse Events (see points a.-e. above) since participant's last assessment/visit?

Please tick: ☐ Yes ☐ No

(If yes, please proceed with the SAE form)

Figure 1 – Flow chart of iCST Serious Adverse Event Reporting Procedure



iCST Serious Adverse Event Reporting Form

PART A (to be completed by Researcher or Principal Investigator)

A1. Completed by: _____

A2. Date form completed: ☐☐ ☐☐ ☐☐☐☐ (d d m m y y y y)

A3. Participant Identity (Trial) Number ☐☐☐☐☐☐

A4. How did you become aware of this incident?

_____

A5. Was this SAE suffered by the participant or carer? *Please place an "x" in* **one** *box only.*

Participant ☐
Carer ☐

A6. Are you reporting a death? *Please place an "x" in* **one** *box only.*

Yes ☐ Please proceed to **Question A8**
No ☐ Please proceed to **Question A7**

A7. Please categorise this event, by placing an "x" in all appropriate options.

☐ Life threatening
☐ Hospitalisation or prolongation of existing hospitalisation
☐ Persistent or significant disability or incapacity
☐ Otherwise considered medically significant by the investigator
☐ Alleged/suspected abuse/neglect, as detailed in protection of vulnerable adults protocol

A8. Date of SAE: ☐☐ ☐☐ ☐☐☐☐ (d d m m y y y y)

A9. Location of SAE: _____

A10. Describe the circumstances of the event. Is there any evidence that participation in the trial may have been a contributing factor?

(Attach further sheets if necessary)

PART B (to be completed by Principal Investigator)

B1. In your opinion, did this SAE arise as a result of the participant's or caregiver's involvement in the iCST trial? *Please place an "x" in* **one** *box only.*

Yes ☐
No ☐

B2. Please add any comments regarding the SAE.

Please complete the details below:

B3. Name of Researcher / PI: _____

Please send an electronic version to iracema.leroi@manchester.ac.uk, sheree.mccormick@manchester.ac.uk and susan.dobson@mhsc.nhs.uk .

B4. Signature of Researcher / PI:

B5. Date of signature: ☐☐ / ☐☐ / ☐☐☐☐ (d d / m m / y y y y)

Please print two copies. After signature, please send by post to the address below and retain a copy for the Investigator's Site File.

Dr. Iracema Leroi,
3rd Floor, Jean McFarlane Building,
University of Manchester,
Oxford Road,
Manchester.
M13 9L

PART C (to be completed by Chief Investigator)

C1. Action taken:

C2. Name of CI: Dr. Iracema Leroi

C3. Signature of CI:

C4. Date of signature: ☐☐ / ☐☐ / ☐☐☐☐ (d d / m m / y y y y)

Research Incident Reporting Form

Study:	
Chief Investigator:	**Participant ID**
Principal Investigator:	**Date of Incident:**
Description of Incident:	
Impact to the trial (actual and/or potential):	
Action taken (A)at site, B)by Sponsor C) in response to the event D) to prevent reoccurrence:	
Date PI Informed:	**Date Sponsor Representative Informed (if appropriate):**
Date Incident Resolved:	
Name and role of person reporting Research Incident:	
Signature:	**Date:**

Protocol Deviation Log

Study: INVEST

Chief Investigator: Dr Iracema Leroi **Principal Investigator:**

	Date of protocol deviation	Date Identified	Site/ researcher/ Participant ID	Details of protocol deviation	Date deviation details sent to Trial Manager	Corrective and preventative actions taken to prevent reoccurrence OR no resolution required
1						

Protocol Violation Log

Study: INVEST

Chief Investigator: Dr Iracema Leroi **Principal Investigator:**

	Date of protocol violation	Site/ researcher / Participant ID	Details of protocol violation including actual and potential consequences	Impact to participant / study outcome	Date violation details sent to Trial Manager	Date violation sent to Sponsor Representative	Resolution A) at Site B) by Sponsor C) in response to the event D) to prevent reoccurrence
1							

Participant ID ☐

Withdrawal form

TO BE COMPLETED FOLLOWING A REQUEST FOR WITHDRAWAL

Date of withdrawal request (dd/mm/yyyy): ☐☐☐☐☐☐☐☐

Who has requested the withdrawal?

- ☐ Participant
- ☐ Companion or personal consultee
- ☐ Nominated consultee

Please tick the phase when the withdrawal was requested:

- ☐ Informed consent
- ☐ Companion training
- ☐ Baseline assessments
- ☐ During the 10 week trial
- ☐ Follow-up assessments
- ☐ Post-therapy interviews
- ☐ Post-therapy focus groups
- ☐ During the extension (+10 week) trial
- ☐ Other (please state): _____

Has the participant/caregiver given a reason for withdrawal? ☐ Yes ☐ No

If yes, please provide details: .....

.....

To be completed by the researcher

Participant Identity Number ☐

Completed by (please print name): _____

Signed: ☐

Date (dd/mm/yyyy): ☐☐☐☐☐☐☐☐

Appendix D: Assessments

Participant ID

Participant Questionnaire during consent I

This sheet should be completed by project researchers following consent with participants and companions during first study visit.

Instructions for the interviewer

Before commencing the interview, please insert the Participant Identity Number on the questionnaire booklet (using the boxes below).

To ensure the smooth operation of the data management, it would be appreciated if the following could be observed:

- Please complete the form using a black or blue pen
- Please do not fold or crease the form
- Please complete all the questions if possible
- Please enter your responses in the boxes/spaces provided, as instructed
- Please use only a single line to delete mistakes and initial each such correction

At the end of the interview please complete the boxes below.

Thank you for your cooperation!

To be completed by the interviewer

Participant Identity Number

Clinic, hospital or other where participant first contacted: _____

Completed by (please print name): _____

Signed:

Interview date (dd/mm/yyyy):

Participant ID ☐

Sociodemographic Questionnaire: Background details

This section is completed by interviewing the companion.

Provide the companion with the following information: To help with our study it will be helpful to have some background details about you and your relative. These will allow us to compare groups in the study with the general population. All data is confidential and stored in an anonymised form.

1. What is your relationship to the participant?
Please tick one box only

Wife/Husband (spouse)	☐	Brother/sister	☐
Partner	☐	Other relative	☐
Son/daughter	☐	Friend	☐
Son/daughter-in-law	☐		
Other (please specify)	☐	.....	

2. How long have you known each other? ..... years

3. How long have you been caring for the person (if applicable)? ..... years

4. Approximately how many hours per week do you spend on caring? ..... hours

5. Please indicate the gender of the participant and companion
Tick as appropriate

Participant	Male	☐	Female	☐
Companion	Male	☐	Female	☐

6. Please indicate the marital status of the participant and companion
Please tick one box only for each

Participant	**Companion**
☐ Single (never married)	☐ Single (never married)
☐ Married	☐ Married
☐ Co-habiting	☐ Co-habiting
☐ Civil partnership	☐ Civil partnership
☐ Separated	☐ Separated
☐ Divorced	☐ Divorced
☐ Widowed	☐ Widowed

7. Please indicate the living status of the participant and companion *(please tick all that apply)*

Participant lives with

- ☐ Spouse/Partner
- ☐ Other family
- ☐ Other
- ☐ No-one

Companion lives with

- ☐ Spouse/Partner
- ☐ Other family
- ☐ Other
- ☐ No-one

8. Ethnicity of the participant and companion
Please tick one box only for each

Participant

WHITE	☐ White British ☐ White Irish ☐ Other White background
BLACK or BLACK BRITISH	☐ Caribbean ☐ African ☐ Other Black background
MIXED	☐ White & Black Caribbean ☐ White & Black African ☐ White & Asian ☐ Other Mixed Background
ASIAN or ASIAN BRITISH	☐ Indian ☐ Pakistani ☐ Bangladeshi ☐ Other Asian background
CHINESE	☐ Chinese
OTHER	☐ Other ethnic group ☐ Not stated ☐ Do not wish to specify

Companion

WHITE	☐ White British ☐ White Irish ☐ Other White background
BLACK or BLACK BRITISH	☐ Caribbean ☐ African ☐ Other Black background
MIXED	☐ White & Black Caribbean ☐ White & Black African ☐ White & Asian ☐ Other Mixed Background
ASIAN or ASIAN BRITISH	☐ Indian ☐ Pakistani ☐ Bangladeshi ☐ Other Asian background
CHINESE	☐ Chinese
OTHER	☐ Other ethnic group ☐ Not stated ☐ Do not wish to specify

9. At what age did the participant and companion leave full-time education?

Participant ☐☐☐ **Not applicable** (i.e. never had formal education) ☐

Companion ☐☐☐ **Not applicable** (i.e. never had formal education) ☐

10. Please indicate the highest level of education of the participant and companion
Please tick one box only

Participant:

- ☐ School Leaver (14-16 years of age)
- ☐ School Leaver (18 years of age)
- ☐ Further Education (Vocational qualifications: i.e. GNVQ/NVQ/HND)
- ☐ Higher Education (BSc/BA or equivalent)
- ☐ Postgraduate Education (MSc/MA/PhD or equivalent)

Companion:

- ☐ School Leaver (14-16 years of age)
- ☐ School Leaver (18 years of age)
- ☐ Further Education (Vocational qualifications: i.e. GNVQ/NVQ/HND)
- ☐ Higher Education (BSc/BA or equivalent)
- ☐ Postgraduate Education (MSc/MA/PhD or equivalent)

Participant ID ☐

11. Is the participant retired? ☐ Yes ☐ No *(proceed to 12c)*

12a. If yes, what was the participant's occupation prior to retirement?

.....

12b. If yes, what was the reason for retirement?

.....

12c. If not, what is the participant's current occupation?

.....

13. Is the companion retired? ☐ Yes ☐ No *(proceed to 14c)*

14a. If yes, what was the companion's occupation prior to retirement?

.....

14b. If yes, what was the reason for retirement?

.....

14c. If not, what is the companion's current occupation?

.....

15. Does the participant have any hearing impairments? ☐ Yes ☐ No

15a. If yes, does the participant use a hearing aid? ☐ Yes ☐ No

16a. Does the participant have any visual impairments? *Please tick one box only*

- ☐ None
- ☐ Partially sighted
- ☐ Wears glasses
- ☐ Other *(please specify)*: .....

16b. If yes, does the participant have any difficulty reading a newspaper? ☐ Yes ☐ No

16c. If yes, does the participant experience any difficulty looking at coloured photos?

☐ Yes ☐ No

17. Please complete the following questions with the participant (with the help from companion if required). If yes was ticked for any questions, please add frequency per week.

Questions	Yes/No	If yes, how many times per week?
17a. Do you maintain an active social life (outside of the house)?		
17b. Do you engage in hobbies (reading, puzzles, crosswords)?		
17c. Do you engage in sports/physical activity?		
17d. Do you follow current affairs (TV/news)?		
17e. Does your family/friends come over to visit?		
17f. Do you reminisce with family/friends about the past?		

PLEASE COMPLETE PARTICIPANT IDENTIFICATION FORM (including GP details)

PARKINSON'S DISEASE

18. Confirmed diagnosis (*please write*): .....

19. Year of PD/DLB diagnosis: ..... (year) ..... (month)

20. Onset of motor symptoms: ..... (year) ..... (month)

21. First motor symptoms of the participant (*Please tick all that apply*):

- ☐ Tremor (including internal tremor)
- ☐ Stiffness
- ☐ Change in facial expression
- ☐ Disturbances of dexterity
- ☐ Micrographia
- ☐ Weakness
- ☐ Dystonia
- ☐ Freezing
- ☐ Lack of arm swing
- ☐ Leg dragging
- ☐ Shuffling of gait
- ☐ Postural imbalance (excluding falls)
- ☐ Falls
- ☐ Slowness of gait
- ☐ Other (please specify): .....

21a. Side where the participant's symptoms first presented on: ☐ Left ☐ Right

22. Onset of cognitive problems: ..... (year) ..... (month)

23. Initial cognitive or other non-motor symptoms of the participant *(Please give a brief overview)*:

.....

.....

Past medical history and comorbidities

24. Has the participant ever had a head injury that was not related to the Parkinsonian symptoms? ☐ Yes ☐ No

25. Please list any relevant comorbidities that are not related to PD/DLB?

.....

.....

26. Does the participant have any of the following? (*please tick all that apply*)

☐ Pathological or addictive gambling ☐ Obsessive shopping ☐ Hypersexuality
☐ Binge eating ☐ Punding or compulsive hobbyism ☐ Other .....

27. Please list all the current medication below without dosages:

27a. Medication for Parkinson's disease: .....

.....

27b. Medication for cognitive symptoms: .....

27c. Medication for neuropsychiatric symptoms: .....

27d. Other medication: .....

.....

27e. Timing of medication: .....

NAME :

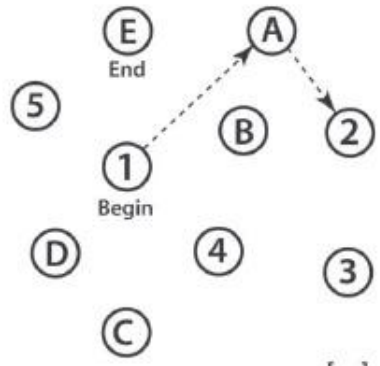
MONTREAL COGNITIVE ASSESSMENT (MOCA)

Version 7.1 Original Version

Education : Date of birth :

Sex : DATE :

VISUOSPATIAL / EXECUTIVE



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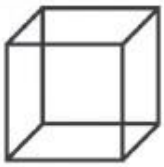


[ ] [ ]

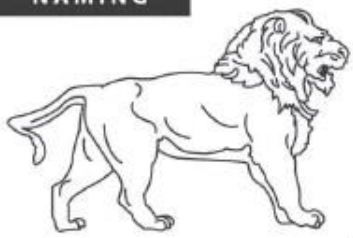
Draw CLOCK (Ten past eleven) (3 points)

[ ] Contour [ ] Numbers [ ] Hands

POINTS

/5

NAMING



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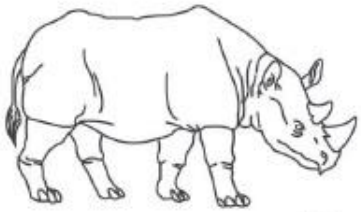

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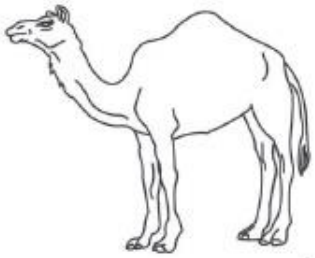


[ ]

/3

MEMORY

Read list of words, subject must repeat them. Do 2 trials, even if 1st trial is successful. Do a recall after 5 minutes.

	FACE	VELVET	CHURCH	DAISY	RED
1st trial					
2nd trial					

No points

ATTENTION

Read list of digits (1 digit/ sec.).

Subject has to repeat them in the forward order [ ] 2 1 8 5 4

Subject has to repeat them in the backward order [ ] 7 4 2

/2

Read list of letters. The subject must tap with his hand at each letter A. No points if ≥ 2 errors

[ ] F B A C M N A A J K L B A F A K D E A A A J A M O F A A B

/1

Serial 7 subtraction starting at 100 [ ] 93 [ ] 86 [ ] 79 [ ] 72 [ ] 65

4 or 5 correct subtractions: **3 pts**, 2 or 3 correct: **2 pts**, 1 correct: **1 pt**, 0 correct: **0 pt**

/3

LANGUAGE

Repeat : I only know that John is the one to help today. [ ]

The cat always hid under the couch when dogs were in the room. [ ]

/2

Fluency / Name maximum number of words in one minute that begin with the letter F [ ] _____ (N ≥ 11 words)

/1

ABSTRACTION

Similarity between e.g. banana - orange = fruit [ ] train – bicycle [ ] watch - ruler

/2

DELAYED RECALL

	Has to recall words WITH NO CUE	FACE [ ]	VELVET [ ]	CHURCH [ ]	DAISY [ ]	RED [ ]	Points for UNCUED recall only
Optional	Category cue						
	Multiple choice cue						

/5

ORIENTATION

[ ] Date [ ] Month [ ] Year [ ] Day [ ] Place [ ] City

/6

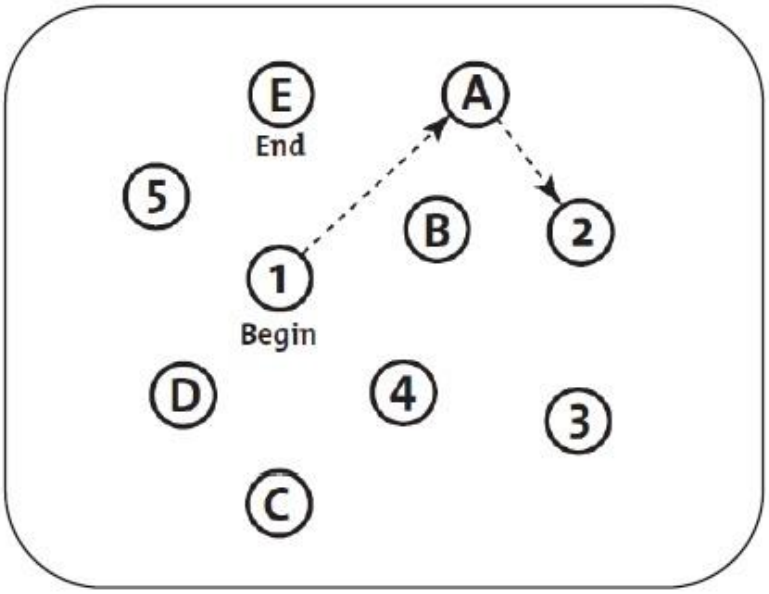
www.mocatest.org

Normal ≥ 26 / 30

TOTAL /30

Add 1 point if ≤ 12 yr edu

Administered by: _____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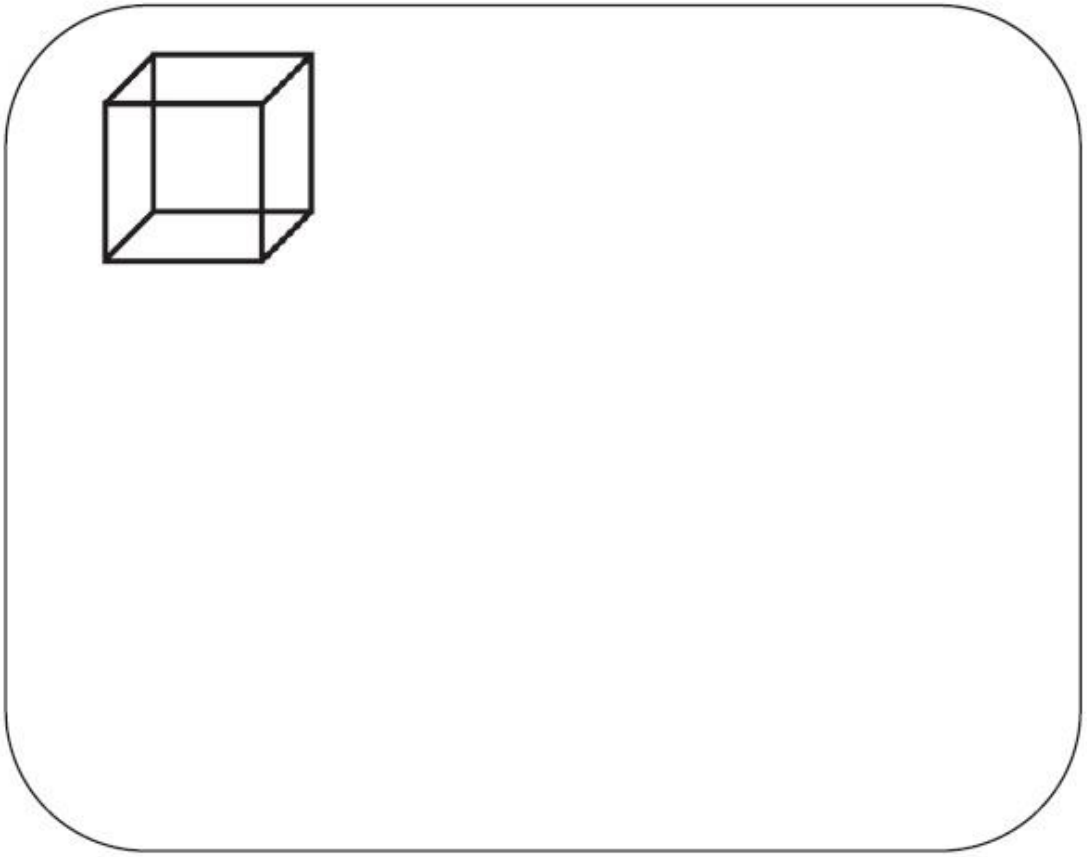
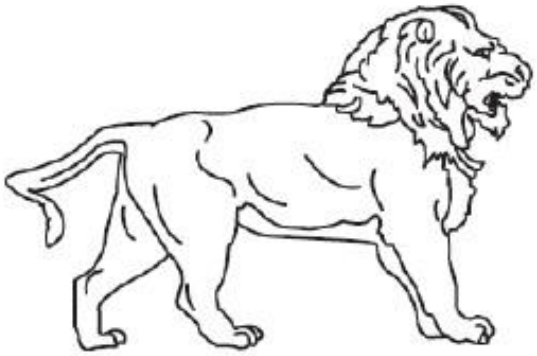
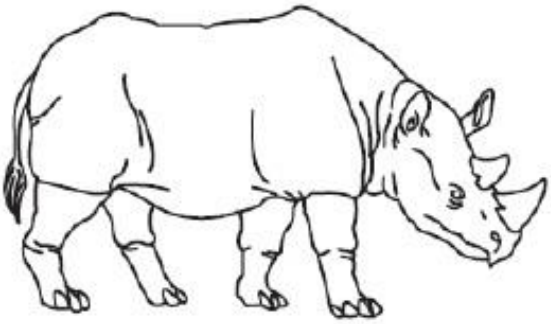
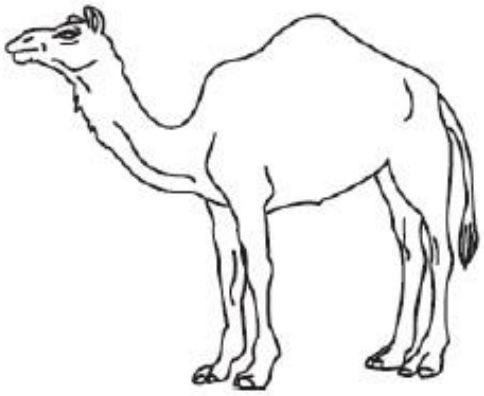
End
A
5
B
2
1
Begin
D
4
3
C







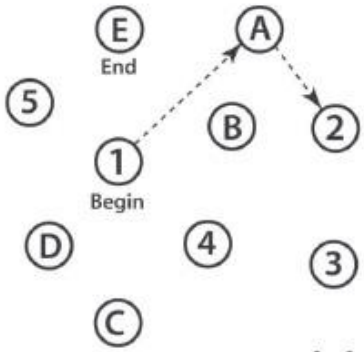


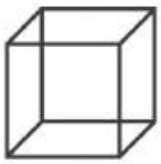
NAME :

MONTREAL COGNITIVE ASSESSMENT (MOCA) Education : Date of birth :

Version 7.1 Original Version Sex : DATE :

VISUOSPATIAL / EXECUTIVE





Copy cube

[ ] [ ]

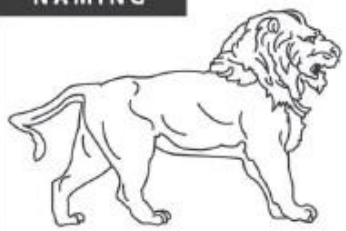
Draw CLOCK (Ten past eleven) (3 points)

[ ] Contour [ ] Numbers [ ] Hands

POINTS

__/5

NAMING



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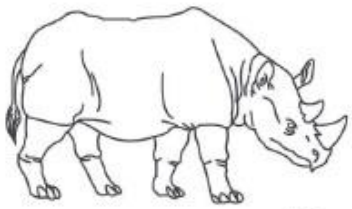

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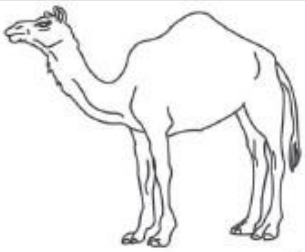

[ ]

__/3

MEMORY Read list of words, subject must repeat them. Do 2 trials, even if 1st trial is successful. Do a recall after 5 minutes.

	FACE	VELVET	CHURCH	DAISY	RED
1st trial					
2nd trial					

No points

ATTENTION Read list of digits (1 digit/ sec.).

Subject has to repeat them in the forward order [ ] 2 1 8 5 4

Subject has to repeat them in the backward order [ ] 7 4 2

__/2

Read list of letters. The subject must tap with his hand at each letter A. No points if ≥ 2 errors

[ ] F B A C M N A A J K L B A F A K D E A A A J A M O F A A B

__/1

Serial 7 subtraction starting at 100 [ ] 93 [ ] 86 [ ] 79 [ ] 72 [ ] 65

4 or 5 correct subtractions: **3 pts**, 2 or 3 correct: **2 pts**, 1 correct: **1 pt**, 0 correct: **0 pt**

__/3

LANGUAGE Repeat : I only know that John is the one to help today. [ ]

The cat always hid under the couch when dogs were in the room. [ ]

__/2

Fluency / Name maximum number of words in one minute that begin with the letter F [ ] _____ (N ≥ 11 words)

__/1

ABSTRACTION Similarity between e.g. banana - orange = fruit [ ] train – bicycle [ ] watch - ruler

__/2

DELAYED RECALL

	Has to recall words WITH NO CUE	FACE	VELVET	CHURCH	DAISY	RED	Points for UNCUED recall only
		[ ]	[ ]	[ ]	[ ]	[ ]	
Optional	Category cue						
	Multiple choice cue						

__/5

ORIENTATION [ ] Date [ ] Month [ ] Year [ ] Day [ ] Place [ ] City

__/6

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Normal ≥ 26 / 30

TOTAL __/30

Add 1 point if ≤ 12 yr edu

Administered by: _____

Unified Parkinson's Disease Rating Scale

Part III – Motor examination

Speech		
Normal	0	
Slight loss of expression, diction or volume	1	
Monotone, slurred but understandable, moderately impaired	2	
Marked impairment, difficult to understand	3	
Unintelligible	4	

Facial expression		
Normal	0	
Minimal hypomimia, could be normal "poker face"	1	
Slight but definitely abnormal diminution of facial expression	2	
Moderate hypomimia, lips parted some of the time	3	
Masked or fixed face, lips parted ¼ inch or more with complete loss of expression	4	

Tremor at rest (hands placed on the arms of the chair)		Face	RUE	LUE	RLE	LLE
Absent	0					
Slight and infrequently present	1					
Mild in amplitude and persistent. Or moderate in amplitude, but only intermittently present	2					
Moderate in amplitude and present most of the time	3					
Marked in amplitude and present most of the time	4					

Action or postural tremor of hands (3 finger-to-nose movements with each hand)		RUE	LUE
Absent	0		
Slight, present with action	1		
Moderate in amplitude, present with action	2		
Moderate in amplitude with posture holding as well as action	3		
Marked in amplitude, interferes with feeding	4		

Finger taps (patient taps thumb with index finger in rapid succession)		RUE	LUE
Normal	0		
Mild slowing and/or reduction in amplitude	1		
Moderately impaired, definite and early fatiguing. May have occasional arrests in movement	2		
Severely impaired, frequent hesitation in initiating movements or arrests in ongoing movement	3		
Can barely perform the task	4		

Hand movements (patient opens and closes hands in rapid succession 10x, from tight fist with the arm bent at the elbow so that the palm faces the rater)		RUE	LUE
Normal	0		
Mild slowing and/or reduction in amplitude	1		
Moderately impaired, definite and early fatiguing. May have occasional arrests in movement	2		
Severely impaired, frequent hesitation in initiating movements or arrests in ongoing movement	3		
Can barely perform the task	4		

Rapid alternating movements of hands (pronation-supination movements of hands, vertically and horizontally, with as large an amplitude as possible, both hands simultaneously) arms extended in front with the palms down, then turn the palm up and down 10x (fast & fully)		RUE	LUE
Normal	0		
Mild slowing and/or reduction in amplitude	1		
Moderately impaired, definite and early fatiguing. May have occasional arrests in movement	2		
Severely impaired, frequent hesitation in initiating movements or arrests in ongoing movement	3		
Can barely perform the task	4		

Leg agility (patient taps heel on the ground in rapid succession picking up entire leg, amplitude should be at least 3 inches)		RLE	LLE
Normal	0		
Mild slowing and/or reduction in amplitude	1		
Moderately impaired, definite and early fatiguing. May have occasional arrests in movement	2		
Severely impaired, frequent hesitation in initiating movements or arrests in ongoing movement	3		
Can barely perform the task	4		

Rigidity – muscle stiffness (judged on passive movement of joints with patient relaxed in a sitting position) First test without an activation manoeuvre. If no rigidity is detected use an activation manoeuvre such as tapping hand on their other knee.	Neck	RUE	LUE	RLE	LLE

Rigidity		Neck	RUE	LUE	RLE	LLE
Absent	0					
Slight or detectable only when activated by mirror or other movements	1					
Mild to moderate	2					
Marked, but full range of motion easily achieved	3					
Severe, range of motion achieved with difficulty	4					

Arising from chair (patient attempts to rise from a straight-backed chair, with arms folded across chest)		
Normal	0	
Slow, or may need more than one attempt	1	
Pushes self up from arms of seat	2	
Tends to fall back and may have to try more than one time, but can get up without help	3	
Unable to arise without help	4	

Posture (assessed when standing and walking, rate the worst posture seen)		
Normal	0	
Not quite erect, slightly stooped posture; could be normal for older person	1	
Moderately stooped posture, definitely abnormal; can be slightly leaning to one side	2	
Severely stooped posture with kyphosis; can be moderately leaning to one side	3	
Marked flexion with extreme abnormality of posture	4	

Gait (have the patient walk away from and towards the examiner to assess both sides. They should walk 10m/30ft, then turn around and return.) festination = small quick steps		
Normal	0	
Walks slowly, may shuffle with short steps, but no propulsion or festination	1	
Walks with difficulty, but requires little or no assistance; may have some festination, short steps or propulsion	2	
Severe disturbance of gait, requiring assistance	3	
Cannot walk at all, even with assistance	4	

Postural stability (response to sudden, strong posterior displacement produced by pull on shoulders while patient erect with eyes open and feet slightly apart. Patient is prepared)		
Normal (no problems, recovers with 1 or 2 steps)	0	
Retropulsion, but recovers unaided (3-5 steps)	1	
Absence of postural response; would fall if not caught by examiner (5+ steps)	2	
Very unstable, tends to lose balance spontaneously (stands safely, but with absence of postural response; falls if not caught by examiner)	3	
Unable to stand without assistance (very unstable, loses balance easily)	4	

Body bradykinesia and hypokinesia (combining slowness, hesitancy, decreased armswing, small amplitude, and poverty of movement in general)		
Normal	0	
Minimal slowness, giving movement a deliberate character; could be normal for some persons. Possibly reduced amplitude	1	
Mild degree of slowness and poverty of movement which is definitely abnormal. Alternatively, some reduced amplitude	2	
Moderate slowness, poverty or small amplitude of movement	3	
Marked slowness, poverty or small amplitude of movement	4	

Total score part III	

V. Modified Hoehn and Yahr Scale

Unilateral symptoms only	1.0	
Unilateral and axial involvement	1.5	
Bilateral symptoms, no impairment of balance	2.0	
Mild bilateral disease with recovery on the pull test	2.5	
Mild-moderate bilateral disease; some postural instability; physically independent	3.0	
Severe disability; still able to walk or stand unaided	4.0	
Wheelchair-bound or bedridden unless aided	5.0	

VI. Schwab and England Activities of Daily Scale

100%	Completely independent. Able to do all chores without slowness, difficulty or impairment. Essentially normal. Unaware of any difficulty.
90%	Completely independent. Able to do all chores with some degree of slowness, difficulty and impairment. Might take twice as long. Beginning to be aware of difficulty.
80%	Completely independent in most chores. Takes twice as long. Conscious of difficulty and slowness.
70%	Not completely independent. More difficulty with some chores. Three to four times as long in some. Must spend a large part of the day with chores.
60%	Some dependency. Can do most chores, but exceedingly slowly and with much effort. Errors; some impossible.
50%	More dependent. Help with ½ of chores, slower, etc. Difficulty with everything.
40%	Very dependent. Can assist with all chores, but few alone.
30%	With effort, now and then does a few chores alone or begins alone. Much help needed.
20%	Nothing alone. Can be a slight help with some chores. Severe invalid.
10%	Totally dependent, helpless. Complete invalid.
0%	Vegetative functions such as swallowing, bladder and bowel functions are not functioning. Bedridden.

Proxy-completed Questionnaire (Pack III)

A. DELUSIONS **(NA)**

Does the patient have beliefs that you know are not true (for example, insisting that people are trying to harm him/her or steal from him/her)? Has he/she said that family members are not who they say they are or that the house is not their home? I'm not asking about mere suspiciousness; I am interested if the patient is convinced that these things are happening to him/her.

☐ Yes (If yes, please proceed to subquestions)
☐ No (if no, please proceed to next screening question) ☐ N/A

1. Does the patient believe that he/she is in danger - that others are planning to hurt him/her?	☐ Yes	☐ No
2. Does the patient believe that others are stealing from him/her?	☐ Yes	☐ No
3. Does the patient believe that his/her spouse is having an affair?	☐ Yes	☐ No
4. Does the patient believe that unwelcome guests are living in his/her house?	☐ Yes	☐ No
5. Does the patient believe that his/her spouse or others are not who they claim to be?	☐ Yes	☐ No
6. Does the patient believe that his/her house is not his/her home?	☐ Yes	☐ No
7. Does the patient believe that family members plan to abandon him/her?	☐ Yes	☐ No
8. Does the patient believe that television or magazine figures are actually present in the home? (Does he/she try to talk or interact with them?)	☐ Yes	☐ No
9. Does the patient believe any other unusual things that I haven't asked about?	☐ Yes	☐ No

If the screening question is confirmed, determine the frequency and severity of the delusions.

Frequency:

☐ 1. Rarely – less than once per week
☐ 2. Sometimes – about once per week
☐ 3. Often – several times per week but less than every day
☐ 4. Very often – once or more per day

Severity:

☐ 1. Mild – delusions present but seem harmless and produce little distress in the patient.
☐ 2. Moderate – delusions are distressing and disruptive.
☐ 3. Severe – delusions are very disruptive and are a major source of behavioral disruption. (If PRN medications are prescribed, their use signals that the delusions are of marked severity.)

Distress: How emotionally distressing do you find this behavior?

☐ 0. Not at all
☐ 1. Minimally (almost no change in work routine)
☐ 2. Mildly (almost no change in work routine but little time rebudgeting required)
☐ 3. Moderately (disrupts work routine, requires time rebudgeting)
☐ 4. Severely (disruptive, upsetting to staff and other residents, major time infringement)
☐ 5. Very Severely or Extremely (very disruptive, major source of distress for staff and other residents, requires time usually devoted to other residents or activities)

8

B. HALLUCINAIONS **(NA)**

Does the patient have hallucinations such as seeing false visions or hearing false voices? Does he/she seem to see, hear or experience things that are not present? By this question we do not mean just mistaken beliefs such as stating that someone who has died is still alive; rather we are asking if the patient actually has abnormal experiences of sounds or visions.

☐ Yes (if yes, please proceed to subquestions)
☐ No (if no, please proceed to next screening question) ☐ N/A

1. Does the patient describe hearing voices or act as if he/she hears voices?	☐ Yes	☐ No
2. Does the patient talk to people who are not there?	☐ Yes	☐ No
3. Does he/she describe seeing things not seen by others or behave as if he/she is seeing things not seen by others (people, animals, lights, etc)?	☐ Yes	☐ No
4. Does he/she report smelling odors not smelled by others?	☐ Yes	☐ No
5. Does he/she describe feeling things on his/her skin or otherwise appear to be feeling things crawling or touching him/her?	☐ Yes	☐ No
6. Does he/she describe tastes that are without any known cause?	☐ Yes	☐ No
7. Does he/she describe any other unusual sensory experiences?	☐ Yes	☐ No

If the screening question is confirmed, determine the frequency and severity of the hallucinations.

Frequency:

☐ 1. Rarely – less than once per week.

☐ 2. Sometimes – about once per week.

☐ 3. Often – several times per week but less than every day.

☐ 4. Very often – once or more per day.

Severity:

☐ 1. Mild – hallucinations are present but harmless and cause little distress for the patient.

☐ 2. Moderate – hallucinations are distressing and are disruptive to the patient.

☐ 3. Severe – hallucinations are very disruptive and are a major source of behavioral disturbance. PRN medications may be required to control them.

Distress: How emotionally distressing do you find this behavior?

☐ 0. Not at all

☐ 1. Minimally (almost no change in work routine)

☐ 2. Mildly (almost no change in work routine but little time rebudgeting required)

☐ 3. Moderately (disrupts work routine, requires time rebudgeting)

☐ 4. Severely (disruptive, upsetting to staff and other residents, major time infringement)

☐ 5. Very Severely or Extremely (very disruptive, major source of distress for staff and other residents, requires time usually devoted to other residents or activities)

9

C. AGITATION/AGGRESSION (NA)

Does the patient have periods when he/she refuses to cooperate or won't let people help him/her? Is he/she hard to handle?

☐ Yes (if yes, please proceed to subquestions)
☐ No (if no, please proceed to next screening question) ☐ N/A

1. Does the patient get upset with those trying to care for him/her or resist activities such as bathing or changing clothes?	☐ Yes	☐ No
2. Is the patient stubborn, having to have things his/her way?	☐ Yes	☐ No
3. Is the patient uncooperative, resistive to help from others?	☐ Yes	☐ No
4. Does the patient have any other behaviors that make him/her hard to handle?	☐ Yes	☐ No
5. Does the patient shout or curse angrily?	☐ Yes	☐ No
6. Does the patient slam doors, kick furniture, throw things?	☐ Yes	☐ No
7. Does the patient attempt to hurt or hit others?	☐ Yes	☐ No
8. Does the patient have any other aggressive or agitated behaviors?	☐ Yes	☐ No

If the screening question is confirmed, determine the frequency and severity of the agitation/aggression.

Frequency:

☐ 1. Rarely – less than once per week.
☐ 2. Sometimes – about once per week.
☐ 3. Often – several times per week but less than every day.
☐ 4. Very often – once or more per day.

Severity:

☐ 1. Mild – agitation is disruptive but can be managed with redirection or reassurance.
☐ 2. Moderate – agitation is disruptive and difficult to redirect or control.
☐ 3. Severe – agitation is very disruptive and a major source of difficulty; there may be a threat of personal harm. Medications are often required.

Distress: How emotionally distressing do you find this behavior?

☐ 0. Not at all
☐ 1. Minimally (almost no change in work routine)
☐ 2. Mildly (almost no change in work routine but little time rebudgeting required)
☐ 3. Moderately (disrupts work routine, requires time rebudgeting)
☐ 4. Severely (disruptive, upsetting to staff and other residents, major time infringement)
☐ 5. Very Severely or Extremely (very disruptive, major source of distress for staff and other residents, requires time usually devoted to other residents or activities)

10

D. DEPRESSION/DYSPHORIA (NA)

Does the patient seem sad or depressed? Does he/she say that he/she feels sad or depressed?

☐ Yes (if yes, please proceed to subquestions)
☐ No (if no, please proceed to next screening question) ☐ N/A

1. Does the patient have periods of tearfulness or sobbing that seem to indicate sadness?	☐ Yes	☐ No
2. Does the patient say, or act as if, he/she is sad or in low spirits?	☐ Yes	☐ No
3. Does the patient put him/herself down or say that he/she feels like a failure?	☐ Yes	☐ No
4. Does the patient say that he/she is a bad person or deserves to be punished?	☐ Yes	☐ No
5. Does the patient seem very discouraged or say that he/she has no future?	☐ Yes	☐ No
6. Does the patient say he/she is a burden to the family or that the family would be better off without him/her?	☐ Yes	☐ No
7. Does the patient express a wish for death or talk about killing himself/herself?	☐ Yes	☐ No
8. Does the patient show any other signs of depression or sadness?	☐ Yes	☐ No

If the screening question is confirmed, determine the frequency and severity of the depression/dysphoria.

Frequency:

☐ 1. Rarely – less than once per week.

☐ 2. Sometimes – about once per week.

☐ 3. Often – several times per week but less than every day.

☐ 4. Very often – essentially continuously present.

Severity:

☐ 1. Mild – depression is distressing but usually responds to redirection or reassurance.

☐ 2. Moderate – depression is distressing; depressive symptoms are spontaneously voiced by the patient and difficult to alleviate.

☐ 3. Severe – depression is very distressing and a major source of suffering for the patient.

Distress: How emotionally distressing do you find this behavior?

☐ 0. Not at all

☐ 1. Minimally (almost no change in work routine)

☐ 2. Mildly (almost no change in work routine but little time rebudgeting required)

☐ 3. Moderately (disrupts work routine, requires time rebudgeting)

☐ 4. Severely (disruptive, upsetting to staff and other residents, major time infringement)

☐ 5. Very Severely or Extremely (very disruptive, major source of distress for staff and other residents, requires time usually devoted to other residents or activities)

11

E. ANXIETY (NA)

Is the patient very nervous, worried, or frightened for no apparent reason? Does he/she seem very tense or fidgety? Is the patient afraid to be apart from you?

☐ Yes (if yes, please proceed to subquestions)
☐ No (if no, please proceed to next screening question) ☐ N/A

1. Does the patient say that he/she is worried about planned events?	☐ Yes	☐ No
2. Does the patient have periods of feeling shaky, unable to relax, or feeling excessively tense?	☐ Yes	☐ No
3. Does the patient have periods of [or complain of] shortness of breath, gasping, or sighing for no apparent reason other than nervousness?	☐ Yes	☐ No
4. Does the patient complain of butterflies in his/her stomach, or of racing or pounding of the heart in association with nervousness? (Symptoms not explained by ill health)	☐ Yes	☐ No
5. Does the patient avoid certain places or situations that make him/her more nervous such as riding in the car, meeting with friends, or being in crowds?	☐ Yes	☐ No
6. Does the patient become nervous and upset when separated from you (or his/her caregiver)? (Does he/she cling to you to keep from being separated?)	☐ Yes	☐ No
7. Does the patient show any other signs of anxiety?	☐ Yes	☐ No

If the screening question is confirmed, determine the frequency and severity of the anxiety.

Frequency:

☐ 1. Rarely – less than once per week.

☐ 2. Sometimes – about once per week.

☐ 3. Often – several times per week but less than every day.

☐ 4. Very often – once or more per day.

Severity:

☐ 1. Mild –anxiety is distressing but usually responds to redirection or reassurance.

☐ 2. Moderate – anxiety is distressing, anxiety symptoms are spontaneously voiced by the patient and difficult to alleviate.

☐ 3. Severe – anxiety is very distressing and a major source of suffering for the patient.

Distress: How emotionally distressing do you find this behavior?

☐ 0. Not at all

☐ 1. Minimally (almost no change in work routine)

☐ 2. Mildly (almost no change in work routine but little time rebudgeting required)

☐ 3. Moderately (disrupts work routine, requires time rebudgeting)

☐ 4. Severely (disruptive, upsetting to staff and other residents, major time infringement)

☐ 5. Very Severely or Extremely (very disruptive, major source of distress for staff and other residents, requires time usually devoted to other residents or activities)

12

F. ELATION/EUPHORIA (NA)

Does the patient seem too cheerful or too happy for no reason? I don't mean the normal happiness that comes from seeing friends, receiving presents, or spending time with family members. I am asking if the patient has a persistent and abnormally good mood or finds humor where others do not.

☐ Yes (if yes, please proceed to subquestions)
☐ No (if no, please proceed to next screening question) ☐ N/A

1. Does the patient appear to feel too good or to be too happy, different from his/her usual self?	☐ Yes	☐ No
2. Does the patient find humor and laugh at things that others do not find funny?	☐ Yes	☐ No
3. Does the patient seem to have a childish sense of humor with a tendency to giggle or laugh inappropriately (such as when something unfortunate happens to others)?	☐ Yes	☐ No
4. Does the patient tell jokes or make remarks that are not funny to others but seem funny to him/her?	☐ Yes	☐ No
5. Does he/she play childish pranks such as pinching or playing "keep away" for the fun of it?	☐ Yes	☐ No
6. Does the patient "talk big" or claim to have more abilities or wealth than is true?	☐ Yes	☐ No
7. Does the patient show any other signs of feeling too good or being too happy?	☐ Yes	☐ No

If the screening question is confirmed, determine the frequency and severity of the elation/euphoria.

Frequency:

☐ 1. Rarely – less than once per week.
☐ 2. Sometimes – about once per week.
☐ 3. Often – several times per week but less than every day.
☐ 4. Very often – essentially continuously present.

Severity:

☐ 1. Mild – elation is notable to friends and family but is not disruptive.
☐ 2. Moderate – elation is notably abnormal.
☐ 3. Severe – elation is very pronounced; patient is euphoric and finds nearly everything to be humorous.

Distress: How emotionally distressing do you find this behavior?

☐ 0. Not at all
☐ 1. Minimally (almost no change in work routine)
☐ 2. Mildly (almost no change in work routine but little time rebudgeting required)
☐ 3. Moderately (disrupts work routine, requires time rebudgeting)
☐ 4. Severely (disruptive, upsetting to staff and other residents, major time infringement)
☐ 5. Very Severely or Extremely (very disruptive, major source of distress for staff and other residents, requires time usually devoted to other residents or activities)

13

G. APATHY/INDIFFERENCE (NA)

Has the patient lost interest in the world around him/her? Has he/she lost interest in doing things or does he/she lack motivation for starting new activities? Is he/she more difficult to engage in conversation or in doing chores? Is the patient apathetic or indifferent?

☐ Yes (if yes, please proceed to subquestions)
☐ No (if no, please proceed to next screening question) ☐ N/A

1. Does the patient seem less spontaneous and less active than usual?	☐ Yes	☐ No
2. Is the patient less likely to initiate a conversation?	☐ Yes	☐ No
3. Is the patient less affectionate or lacking in emotions when compared to his/her usual self?	☐ Yes	☐ No
4. Does the patient contribute less to household chores?	☐ Yes	☐ No
5. Does the patient seem less interested in the activities and plans of others?	☐ Yes	☐ No
6. Has the patient lost interest in friends and family members?	☐ Yes	☐ No
7. Is the patient less enthusiastic about his/her usual interests?	☐ Yes	☐ No
8. Does the patient show any other signs that he/she doesn't care about doing new things?	☐ Yes	☐ No

If the screening question is confirmed, determine the frequency and severity of the apathy/indifference.

Frequency:

☐ 1. Rarely – less than once per week.

☐ 2. Sometimes – about once per week.

☐ 3. Often – several times per week but less than every day.

☐ 4. Very often – nearly always present.

Severity:

☐ 1. Mild – apathy is notable but produces little interference with daily routines; only mildly different from patient's usual behavior; patient responds to suggestions to engage in activities.

☐ 2. Moderate – apathy is very evident; may be overcome by the caregiver with coaxing and encouragement; responds spontaneously only to powerful events such as visits from close relatives or family members.

☐ 3. Severe – apathy is very evident and usually fails to respond to any encouragement or external events.

Distress: How emotionally distressing do you find this behavior?

☐ 0. Not at all

☐ 1. Minimally (almost no change in work routine)

☐ 2. Mildly (almost no change in work routine but little time rebudgeting required)

☐ 3. Moderately (disrupts work routine, requires time rebudgeting)

☐ 4. Severely (disruptive, upsetting to staff and other residents, major time infringement)

☐ 5. Very Severely or Extremely (very disruptive, major source of distress for staff and other residents, requires time usually devoted to other residents or activities)

14

H. DISINHIBITION (NA)

Does the patient seem to act impulsively without thinking? Does he/she do or say things that are not usually done or said in public? Does he/she do things that are embarrassing to you or others?

☐ Yes (if yes, please proceed to subquestions)
☐ No (if no, please proceed to next screening question) ☐ N/A

1. Does the patient act impulsively without appearing to consider the consequences?	☐ Yes	☐ No
2. Does the patient talk to total strangers as if he/she knew them?	☐ Yes	☐ No
3. Does the patient say things to people that are insensitive or hurt their feelings?	☐ Yes	☐ No
4. Does the patient say crude things or make sexual remarks that he/she would not usually have said?	☐ Yes	☐ No
5. Does the patient talk openly about very personal or private matters not usually discussed in public?	☐ Yes	☐ No
6. Does the patient take liberties or touch or hug others in way that is out of character for him/her?	☐ Yes	☐ No
7. Does the patient show any other signs of loss of control of his/her impulses?	☐ Yes	☐ No

If the screening question is confirmed, determine the frequency and severity of the disinhibition.

Frequency:

☐ 1. Rarely – less than once per week.
☐ 2. Sometimes – about once per week.
☐ 3. Often – several times per week but less than every day.
☐ 4. Very often – essentially continuously present.

Severity:

☐ 1. Mild – disinhibition is notable but usually responds to redirection and guidance.
☐ 2. Moderate – disinhibition is very evident and difficult to overcome by the caregiver.
☐ 3. Severe – disinhibition usually fails to respond to any intervention by the caregiver, and is a source of embarrassment or social distress.

Distress: How emotionally distressing do you find this behavior?

☐ 0. Not at all
☐ 1. Minimally (almost no change in work routine)
☐ 2. Mildly (almost no change in work routine but little time rebudgeting required)
☐ 3. Moderately (disrupts work routine, requires time rebudgeting)
☐ 4. Severely (disruptive, upsetting to staff and other residents, major time infringement)
☐ 5. Very Severely or Extremely (very disruptive, major source of distress for staff and other residents, requires time usually devoted to other residents or activities)

15

I. IRRITABILITY/LABILITY (NA)

Does the patient get irritated and easily disturbed? Are his/her moods very changeable? Is he/she abnormally impatient? We do not mean frustration over memory loss or inability to perform usual tasks; we are interested to know if the patient has abnormal irritability, impatience, or rapid emotional changes different from his/her usual self.

☐ Yes (if yes, please proceed to subquestions)
☐ No (if no, please proceed to next screening question) ☐ N/A

1. Does the patient have a bad temper, "flying off the handle" easily over little things?	☐ Yes	☐ No
2. Does the patient rapidly change moods from one to another, being fine one minute and angry the next?	☐ Yes	☐ No
3. Does the patient have sudden flashes of anger?	☐ Yes	☐ No
4. Is the patient impatient, having trouble coping with delays or waiting for planned activities?	☐ Yes	☐ No
5. Is the patient cranky and irritable?	☐ Yes	☐ No
6. Is the patient argumentative and difficult to get along with?	☐ Yes	☐ No
7. Does the patient show any other signs of irritability?	☐ Yes	☐ No

If the screening question is confirmed, determine the frequency and severity of the irritability /lability.

Frequency:

☐ 1. Rarely – less than once per week.
☐ 2. Sometimes – about once per week.
☐ 3. Often – several times per week but less than every day.
☐ 4. Very often – essentially continuously present.

Severity:

☐ 1. Mild – irritability or lability is notable but usually responds to redirection and reassurance.
☐ 2. Moderate – irritability and lability are very evident and difficult to overcome by the caregiver.
☐ 3. Severe – irritability and lability are very evident; they usually fail to respond to any intervention by the caregiver, and they are a major source of distress.

Distress: How emotionally distressing do you find this behavior?

☐ 0. Not at all
☐ 1. Minimally (almost no change in work routine)
☐ 2. Mildly (almost no change in work routine but little time rebudgeting required)
☐ 3. Moderately (disrupts work routine, requires time rebudgeting)
☐ 4. Severely (disruptive, upsetting to staff and other residents, major time infringement)
☐ 5. Very Severely or Extremely (very disruptive, major source of distress for staff and other residents, requires time usually devoted to other residents or activities)

16

J. ABERRANT MOTOR BEHAVIOR (NA)

Does the patient pace, do things over and over such as opening closets or drawers, or repeatedly pick at things or wind string or threads?

☐ Yes (if yes, please proceed to subquestions)
☐ No (if no, please proceed to next screening question) ☐ N/A

1. Does the patient pace around the house without apparent purpose?	☐ Yes	☐ No
2. Does the patient rummage around opening and unpacking drawers or closets?	☐ Yes	☐ No
3. Does the patient repeatedly put on and take off clothing?	☐ Yes	☐ No
4. Does the patient have repetitive activities or "habits" that he/she performs over and over?	☐ Yes	☐ No
5. Does the patient engage in repetitive activities such as handling buttons, picking, wrapping string, etc?	☐ Yes	☐ No
6. Does the patient fidget excessively, seem unable to sit still, or bounce his/her feet or tap his/her fingers a lot?	☐ Yes	☐ No
7. Does the patient do any other activities over and over?	☐ Yes	☐ No

If the screening question is confirmed, determine the frequency and severity of the aberrant motor activity:

Frequency:

☐ 1. Rarely – less than once per week.
☐ 2. Sometimes – about once per week.
☐ 3. Often – several times per week but less than every day.
☐ 4. Very often – essentially continuously present.

Severity:

☐ 1. Mild – abnormal motor activity is notable but produces little interference with daily routines.
☐ 2. Moderate – abnormal motor activity is very evident; can be overcome by the caregiver.
☐ 3. Severe – abnormal motor activity is very evident, usually fails to respond to any intervention by the caregiver and is a major source of distress.

Distress: How emotionally distressing do you find this behavior?

☐ 0. Not at all
☐ 1. Minimally (almost no change in work routine)
☐ 2. Mildly (almost no change in work routine but little time rebudgeting required)
☐ 3. Moderately (disrupts work routine, requires time rebudgeting)
☐ 4. Severely (disruptive, upsetting to staff and other residents, major time infringement)
☐ 5. Very Severely or Extremely (very disruptive, major source of distress for staff and other residents, requires time usually devoted to other residents or activities)

17

Care partner Questionnaire (Pack IV)

INVEST CRF (iCST for PDD/DLB)

Caregiver questionnaire IV
Caregiver ID ☐

Caregiver Questionnaire IV

Thank you for agreeing to participate in this study. In this booklet you will find 9 short questionnaires. Please read the general instructions below before completing the questionnaires, there are no right or wrong answers.

Should you have any difficulties, please ask the visiting researcher for assistance.

General instructions

To ensure the smooth operation of the data management, it would be appreciated if the following could be observed:

- Please complete the form using a black or blue pen
- Please do not fold or crease the form
- Please complete all the questions
- Please enter your responses in the boxes/spaces provided, as instructed
- Please use only a single line to delete mistakes and initial each such correction

Thank you for your cooperation!

To be completed by the interviewer

Is the participant willing to continue taking part in the current study? ☐ Yes ☐ No
If 'No' was ticked, please proceed with a withdrawal request.

Caregiver Identity Number: ☐

Clinic or hospital of first participant contact: _____

Which assessment is this? Please tick **one** box only: Baseline assessment ☐
Follow-up assessment ☐

Completed by (please print name): _____

Which arm do you think the dyad has been randomised to? ☐ Therapy ☐ Control

Signed: ☐

Interview date (dd/mm/yyyy): ☐☐☐☐☐☐☐☐

Questionnaire 1: SF-12® Health Survey

This survey asks for your views about your health. This information will help you keep track of how you feel and how well you are able to do your usual activities.
Answer every question by selecting the answer as indicated. If you are unsure about how to answer a question, please give the best answer you can.

1. In general, would you say your health is:

Excellent	Very good	Good	Fair	Poor
○	○	○	○	○

2. The following questions are about activities you might do during a typical day. Does your health now limit you in these activities? If so, how much?

	Yes, limited a lot	Yes, limited a little	No, not limited at all
a. Moderate activities, such as moving a table, pushing a vacuum cleaner, bowling, or playing golf			
b. Climbing several flights of stairs			

3. During the past 4 weeks, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of your physical health?

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a. Accomplished less than you would like					
b. Were limited in the kind of work or other activities					

4. During the past 4 weeks, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)?

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a. Accomplished less than you would like					
b. Did work or other activities less carefully than usual					

5. During the past 4 weeks, how much did pain interfere with your normal work (including both work outside the home and housework)?

Not at all	A little bit	Moderately	Quite a bit	Extremely
○	○	○	○	○

6. These questions are about how you feel and how things have been with you during the past 4 weeks. For each question, please give the one answer that comes closest to the way you have been feeling.

How much of the time during the past 4 weeks…

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a. Have you felt calm and peaceful?					
b. Did you have a lot of energy?					
c. Have you felt downhearted and blue?					

7. During the past 4 weeks, how much of the time has your physical health or emotional problems interfered with your social activities (like visiting friends, relatives, etc.)?

All of the time	Most of the time	Some of the time	A little of the time	None of the time
○	○	○	○	○

Questionnaire 2: EuroQol EQ-5D

By placing a tick in one box in each group below, please indicate which statements best describe your own health state today.

Mobility
- I have no problems in walking about ☐
- I have some problems in walking about ☐
- I am confined to bed ☐

Self-care
- I have no problems with self-care ☐
- I have some problems with washing or dressing myself ☐
- I am unable to wash or dress myself ☐

Usual activities (*e.g. work, study, housework, family or leisure activities*)
- I have no problems with performing my usual activities ☐
- I have some problems with performing my usual activities ☐
- I am unable to perform my usual activities ☐

Pain/Discomfort
- I have no pain or discomfort ☐
- I have moderate pain or discomfort ☐
- I have extreme pain or discomfort ☐

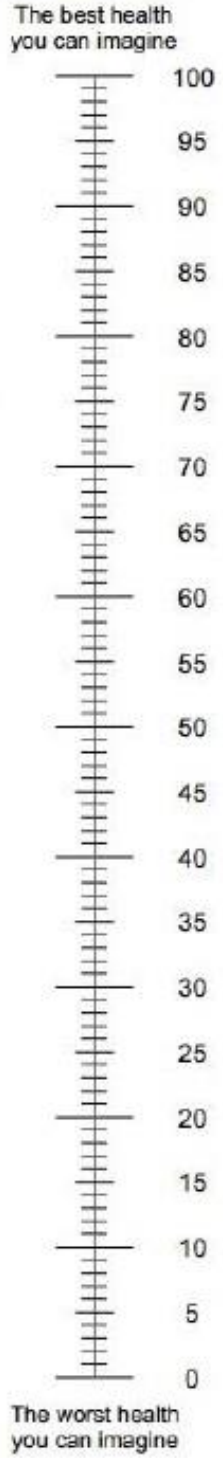
Anxiety/Depression
- I am not anxious or depressed ☐
- I am moderately anxious or depressed ☐
- I am extremely anxious or depressed ☐

EuroQol EQ-5D

To help people say how good or bad a health state is, we have drawn a scale (rather like a thermometer) on which the best state you can imagine is marked 100 and the worst state you can imagine is marked 0.

We would like you to indicate on this scale how good or bad your own health is TODAY, in your opinion. Please do this by drawing a line from the box below to whichever point on the scale indicates how good or bad your health state is today.

Your health state today



Questionnaire 3: Relationship Satisfaction Scale.

Please circle the number on each row that best describes the amount of satisfaction you feel with the person you care for.

	Very dissatisfied	Moderately dissatisfied	Somewhat dissatisfied	Neutral	Somewhat satisfied	Moderately satisfied	Very satisfied
1. Communication and openness	0	1	2	3	4	5	6
2. Resolving conflicts and arguments	0	1	2	3	4	5	6
3. Degree of affection and caring	0	1	2	3	4	5	6
4. Intimacy and closeness	0	1	2	3	4	5	6
5. Satisfaction with your role in the relationship	0	1	2	3	4	5	6
6. Satisfaction with the other person's role in the relationship	0	1	2	3	4	5	6
7. Overall satisfaction with the relationship	0	1	2	3	4	5	6

Questionnaire 4: Brief Resilience Scale.

Please respond to each item by marking one box per row.

	Strongly disagree	Disagree	Neutral	Agree	Strongly agree
1. I tend to bounce back quickly after hard times.	1	2	3	4	5
2. I have a hard time making it through stressful events.	5	4	3	2	1
3. It does not take me long to recover from a stressful event.	1	2	3	4	5
4. It is hard for me to snap back when something bad happens.	5	4	3	2	1
5. I usually come through difficult times with little trouble.	1	2	3	4	5
6. I tend to take a long time to get over set-backs in my life.	5	4	3	2	1

Questionnaire 5: Hospital Anxiety and Depression Scale

Instructions: This questionnaire asks about how you feel. Read each item and circle the reply which comes closest to how you have been feeling in the past week. Don't take too long over your replies: your immediate reaction to each item will probably be more accurate than a long thought out response.

1. I feel tense or 'wound up':	**A**
Most of the time	3
A lot of the time	2
Time to time, occasionally	1
Not at all	0

2. I still enjoy the things I used to enjoy:	**D**
Definitely as much	0
Not quite so much	1
Only a little	2
Not at all	3

3. I get a sort of frightened feeling like something awful is about to happen:	**A**
Very definitely and quite badly	3
Yes, but not too badly	2
A little, but it doesn't worry me	1
Not at all	0

4. I can laugh and see the funny side of things:	**D**
As much as I always could	0
Not quite so much now	1
Definitely not so much now	2
Not at all	3

5. Worrying thoughts go through my mind:	**A**
A great deal of the time	3
A lot of the time	2
From time to time but not too often	1
Only occasionally	0

6. I feel cheerful:	**D**
Not at all	3
Not often	2
Sometimes	1
Most of the time	0

7. I can sit at ease and feel relaxed:	**A**
Definitely	0
Usually	1
Not often	2
Not at all	3

8. I feel as if I am slowed down:	**D**
Nearly all of the time	3
Very often	2
Sometimes	1
Not at all	0

9. I get a sort of frightened feeling like 'butterflies in the stomach':	**A**
Not at all	0
Occasionally	1
Quite often	2
Very often	3

10. I have lost interest in my appearance:	**D**
Definitely	3
I don't take as much care as I should	2
I may not take quite as much care	1
I take just as much care as ever	0

11. I feel restless as if I have to be on the move:	**A**
Very much indeed	3
Quite a lot	2
Not very much	1
Not at all	0

12. I look forward with enjoyment to things:	**D**
A much as I ever did	0
Rather less than I used to	1
Definitely less than I used to	2
Hardly at all	3

13. I get sudden feelings of panic:	**A**
Very often indeed	3
Quite often	2
Not very often	1
Not at all	0

14. I can enjoy a good book or radio or TV programme:	**D**
Often	0
Sometimes	1
Not often	2
Very seldom	3

Questionnaire 6: Zarit Burden Interview.

The following is a list of statements which reflect how people sometimes feel when taking care of another person. After each statement, indicate how often you feel that way: never, rarely, sometimes, quite frequently, or nearly always. There are no right or wrong answers.

	Never	Rarely	Some-times	Quite frequently	Nearly always
1. Do you feel that your relative asks for more help than he or she needs?	0	1	2	3	4
2. Do you feel that, because of the time you spend with your relative, you don't have enough time for yourself?	0	1	2	3	4
3. Do you feel stressed between caring for your relative and trying to meet other responsibilities for your family or work?	0	1	2	3	4
4. Do you feel embarrassed about your relative's behaviour?	0	1	2	3	4
5. Do you feel angry when you are around your relative?	0	1	2	3	4
6. Do you feel that your relative currently affects your relationship with other family members?	0	1	2	3	4
7. Are you afraid about what the future holds for your relative?	0	1	2	3	4
8. Do you feel that your relative is dependent upon you?	0	1	2	3	4
9. Do you feel strained when you are around your relative?	0	1	2	3	4
10. Do you feel that your health has suffered because of your involvement with your relative?	0	1	2	3	4
11. Do you feel that you don't have as much privacy as you would like, because of your relative?	0	1	2	3	4
12. Do you feel that your social life has suffered because you are caring for your relative?	0	1	2	3	4
13. Do you feel uncomfortable having your friends over because of your relative?	0	1	2	3	4
14. Do you feel that your relative seems to expect you to take care of him or her, as if you were the only one he or she could depend on?	0	1	2	3	4
15. Do you feel that you don't have enough money to care for your relative, in addition to the rest of your expenses?	0	1	2	3	4
16. Do you feel that you will be unable to take care of your relative much longer?	0	1	2	3	4
17. Do you feel that you have lost control of your life since your relative's illness?	0	1	2	3	4
18. Do you wish that you could just leave the care of your relative to someone else?	0	1	2	3	4
19. Do you feel uncertain about what to do about your relative?	0	1	2	3	4
20. Do you feel that you should be doing more for your relative?	0	1	2	3	4
21. Do you feel that you could do a better job in caring for your relative?	0	1	2	3	4
22. Overall, how burdened do you feel in caring for your relative?	0	1	2	3	4

Questionnaire 7: Dyadic Relationship Scale. *Please circle the most appropriate answer.*

	Strongly disagree	Disagree	Agree	Strongly agree
1. I felt closer to her/him than I have in awhile.	0	1	2	3
2. I have learned some good things about my (relative).	0	1	2	3
3. I felt angry toward him/her.	3	2	1	0
4. I felt depressed because of my relationship with him/her.	3	2	1	0
5. I felt resentful toward him/her.	3	2	1	0
6. I have had more patience than I have had in the past.	0	1	2	3
7. I have learned some good things about myself.	0	1	2	3
8. I felt that my relationship with him/her was strained.	3	2	1	0
9. I have learned some nice things about other people in my life.	0	1	2	3
10. Communication between my (relative) and me has improved.	0	1	2	3
11. I felt that he/she made requests over and above what he/she needed.	3	2	1	0

Questionnaire 8: Relatives' Stress Scale. *Please circle the most appropriate answer.*

	Never/ not at all	Rarely/ a little	Sometimes/ moderately	Frequently/ quite a lot	Always/ considerably
1. Do you ever feel you can no longer cope with the situation?	0	1	2	3	4
2. Do you ever feel that you need a break?	0	1	2	3	4
3. Do you ever get depressed by the situation?	0	1	2	3	4
4. Has your own health suffered at all?	0	1	2	3	4
5. Do you worry about accident happening to ..... *(your relative or friend)*?	0	1	2	3	4
6. Do you ever feel that there will be no end to the problem?	0	1	2	3	4
7. Do you find it difficult to get away on holiday?	0	1	2	3	4
8. How much has your social life been affected?	0	1	2	3	4
9. How much has the household routine been upset?	0	1	2	3	4
10. Is your sleep interrupted by ..... *(your relative or friend)*?	0	1	2	3	4
11. Has your standard of living been reduced?	0	1	2	3	4
12. Do you ever feel embarrassed by ..... *(your relative or friend)*?	0	1	2	3	4
13. Are you at all prevented from having visitors?	0	1	2	3	4
14. Do you ever get cross and angry with ..... *(your relative or friend)*?	0	1	2	3	4
15. Do you ever feel frustrated at times with ..... *(your relative or friend)*?	0	1	2	3	4

Caregiver questionnaire IV

Caregiver ID

Questionnaire 9: Family caregiving role. *Please circle the most appropriate answer.*

	Strongly disagree	Disagree	Not sure	Agree	Strongly agree
1. I get satisfaction seeing (name) accomplish things.	1	2	3	4	5
2. Caring for (name) has made me more confident dealing with others.	1	2	3	4	5
3. I get a great deal of satisfaction from caring.	1	2	3	4	5
4. I would really feel at a loss if (name) was not around.	1	2	3	4	5
5. I worry about what would happen to (name) if something happened to me.	1	2	3	4	5
6. I feel reassured knowing that as long as I am helping (name) he/she is getting proper care.	1	2	3	4	5
7. I grieve for the opportunities (name) doesn't have.	1	2	3	4	5
8. I have lost control of my life since caring for (name).	1	2	3	4	5
9. I regret the opportunities I don't have.	1	2	3	4	5
10. My friends don't visit as often because I am caring for (name).	1	2	3	4	5
11. I care for (name) because no-one else can.	1	2	3	4	5
12. Caring for (name) takes up most of my time and I neglect the rest of the family.	1	2	3	4	5
13. I am angry when I am around (name).	1	2	3	4	5
14. Nothing I can do seems to please (name).	1	2	3	4	5
15. I am embarrassed over (name's) behaviour.	1	2	3	4	5
16. I feel guilty about (name).	1	2	3	4	5

Thank you for completing the questionnaire!

Your help is much appreciated!

Appendix E: Documents of CST-PD intervention

Topi
1.2

PERSONAL LIFE: MY FAMILY

Family photos and photography

Talk about

- Do you have many photographs from when you were a child, a teenager, a young adult up until now?
- Do you have a favourite photo of yourself, of your family, of your friends? If so, describe the photograph, what is happening on that photograph, what is the emotion like?
- Do you remember a particular time when you were taking photos to capture a particular moment?
- How do our photographs make us feel? Does it create positive memories?
- Why is it good to have photographs?
- How has photography changed over time (e.g. from black and white photos to colourful digital and electronic photos)?
- If you were to take a photo now, what photo or photos would it be? Would it be about a location, a place or nature or about people, friends, family?

Page 7, Personal life – My family

MEDIA & ENTERTAINMENT : TECHNOLOGY

Comparing past and present technology

Look at the images below.

Talk about

- Do you recognise (or own) any of these items?
- When were these items used? Have you used them?
- How are they similar and different to the objects that have since replaced them?

Comparing past and present

- How was data stored? *Compare paper records vs hard-drive.*
- How was music played? *Compare gramophone vs mp3 player.*
- How did people communicate? *Compare a landline phone vs mobile phone. Compare letters by post vs email.*

- Which of these do you prefer?
- How did people communicate before electrical equipment was invented?

TRAVEL & CULTURE: BLACKPOOL, THE HOLIDAY DESTINATION

Blackpool – a popular holiday resort

Talk about

- Describe the images on this page. What are these people doing? How are they feeling?
- What kind of emotions do these images create in you?
- Why do you think Blackpool Tower is presented and visible in so many of these photos? Do you think this might have been to promote Blackpool as a holiday destination?
- How would you describe Blackpool in three words?

Leapfrog

1960s

Think about the 1950s, 60s, 70s, 80s. For each decade, discuss:

- What were popular holiday destinations (or types of holidays)?
- What types of clothes and hairstyles were in fashion?
- What would people of worn on holiday and how would this be different to clothes worn at home?

Topic
7.3

SEASONS: SPRING

Spring

Talk about

- What changes take place in Spring?
- What are some of the spring activities that are done only in the Spring?
- Can you name any celebrations in the Spring season?

Make as many words as you can with the word
SPRING FLOWER

Example: song

- How many words did you get?

Page 166, Seasons – Spring

HOBBIES AND LEISURE: WATER SPORTS

Swimming

Swimming can be a competitive sport for individuals or for teams. In the Olympics the swimmers compete in freestyle, breaststroke, backstroke and butterfly. Can you match the swimming styles with the images?




Talk about

- What style do you think is the fastest? And the slowest?
- What do you need to do in order to win a swimming competition?
- Can you name any benefits of swimming?
- Why knowing how to swim might be a useful skill?
- When do you think is the best age to learn to swim?

GAMES & QUIZZES: MATCH THE PAIRS

Match the pairs

Match each half of the well known pairs with their 'other halves' by drawing a connecting line.

Amounts

Bucket of	Petrol
Pair of	Water/sand
Ball of	Shoes/trousers/glasses
Reel of	Cotton
Gallon of	Bread
Pint of	Water/milk/beer
Jug of	Tea/coffee
Slice of	Wool
Loaf of	Bread/cake/ham
Cup of	Milk/beer

Places

Buckingham	Cross
Westminster	Square
Trafalgar	Hill
Piccadilly	Castle
Nelson's	Abbey
Waterloo	Column
Canterbury	Station
New	Palace
Charing	Square
Times	Circus
Capitol	Cathedral
Windsor	York, Orleans, -castle

Individual Cognitive Stimulation Therapy Workshop

Dr Sheree McCormick (Research Associate)
Sabina Vatter (Research Assistant / PhD Student)

- Division of Neuroscience and Experimental Psychology, University of Manchester.

Workshop Schedule

- Introduction to iCST (15 minutes)
  - Supporting evidence
  - NHS guidelines
- Nine principles of iCST (15 minutes)
- Delivering an iCST session (45 minutes)
  - Trainer to lead a session, caregiver to assist
  - Trainer to lead a session, caregiver to experience the session (differences between effective and non effective sessions)
- Adapting activities/using strategies to overcome challenges (15-30 minutes)
- Questions (15-30 minutes)

Breaks when you need them!

Influencing dementia

- For better (mental stimulation):
  - "Use it or lose it" (Swaab, 1991).
  - Mental activity can lead to new learning and better cognitive functioning.
  - This can involve growth in specific brain areas.
  - (Taxi drivers in London who need to learn The Knowledge.)
- Or for worse (exacerbation):
  - Disempowerment: not allowing someone to use their abilities.
  - Outpacing: acting or behaving at a rate too fast for someone to follow or understand.

Previous therapies

- Reality Orientation (Folson, 1966)
  - One of 1st non-pharmacological interventions. Repetition of information about time, place or person.
- Reminiscence Therapy (Butler and Lewis, 1977)
  - Discussion about the past using pictures, objects, music.
  - Focuses on long-term memory, which is useful in AD.
- Validation therapy (Fell, 1992)
  - Involves telling the life story, with focus on emotions.
  - Emotions cannot be 'right' or 'wrong'.

Cognitive Stimulation Therapy (iCST) for groups (2003)

- 14 sessions (twice a week for seven weeks).
- 201 people with dementia participated.
- Improvements in some memory function and language.
  - "She is brighter."
  - "It relaxed him."
- Therapy was effective whether or not people were on treatment for dementia.
- There was a greater improvement for people on treatment who received therapy than for medication alone.
- Recommended by the National Institute for Health and Clinical Excellent (NICE) for 'all people with mild to moderate dementia.'

iCST for people with movement disorders (INVEST study)

- Includes a number of topics to offer choice/challenges
- Provides opportunity to share enjoyable activities
- Offers caregivers additional/alternative support strategies
- Aims to:
  - Promote confidence and well-being
  - Improve relationships
  - Improve communication

Key Principles

	Key Principles of iCST
1	Considering the person's needs
2	Offering choice
3	Focus on opinions rather than facts
4	Having a tangible focus
5	Using reminiscence
6	Maximising potential
7	Enjoyable and fun
8	Stimulating new ideas and communication
9	Strengthening the caregiving relationship

How will the key principles help me with the therapy?

The key principles are here to help you and your partner to get the most out of the therapy, so it is important that you feel confident and ready to put them into practice. Take some time to read the following pages before you begin, and feel free to revisit them during the therapy. The principles are grouped accorded to when and how you can use them in the session.

1

Putting the key principles into practice

1. Considering the person's needs

We need to see the person first and foremost, rather than focusing on the memory problems and the associated impairments. As you probably know your partner very well you will have a great deal of knowledge about their strengths, think about how you can incorporate their interests in the sessions, rather than concentrating on areas which they might find difficult.

2. Offering choice

The therapy offers a range of different topics and you can tailor the programme to your partner's needs by coming up with your own ideas or using you own resources.

How do I make sure the activities are stimulating?

The brain is like a muscle and has the ability to change through mental practice. A stimulating activity will get your partner thinking and encourage them to explore new ideas. Choose activities that you think will be engaging but take care to ensure the activity is not too difficult so that the person feels deskilled. Mix and match according to your partner's skills and interests.

Problem solving: what if my partner seems to be struggling with the activity I have chosen?

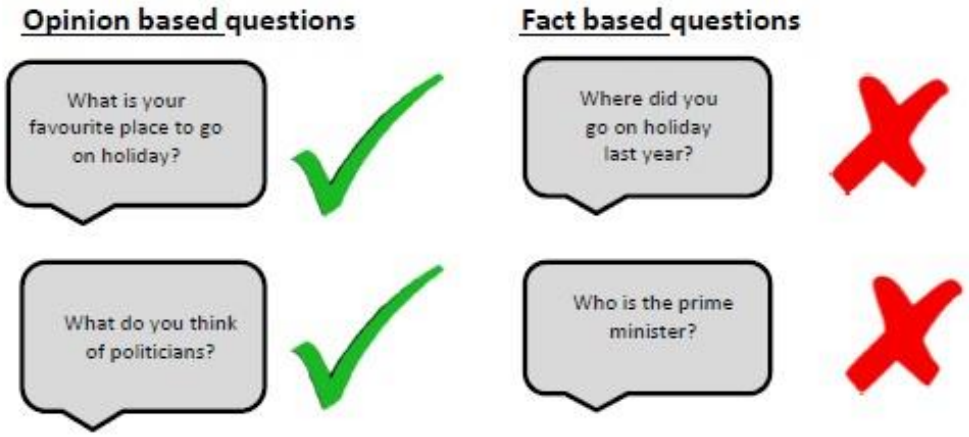
- Some sessions will be more challenging than others, especially since people have different interests and skills.
- If the person asks why things are difficult or seems to be anxious, let them know that you are trying to get them to exercise skills that have not been used for a while, and stimulate different parts of the brain.
- Try an alternative activity if you have time
- If the person is distressed, do not continue with the activity. Try to end on a good note by doing something you know they enjoy instead.

2

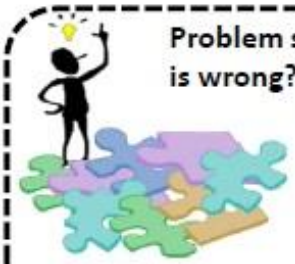
Putting the key principles into practice

3. Opinions rather than facts

In the therapy session we need to focus on the person's strengths. If we focus on facts too much there is the risk that the person might be wrong. If we ask the person for their opinions they cannot be wrong as opinions are neither right or wrong. At first making sure you ask questions in this way might feel challenging, but with practice it will become second nature. Here are some examples:



- Use the manual to jot down some opinion based questions and have them ready during the session.
- If there is a fact based element to the activity, give the person a selection of options to choose from or cues such as images from the manual to help them find the answer.



Problem solving: what if my partner says something that I know is wrong?

- It doesn't matter if your partner offers comments that you know are not right, there is no need to correct them, just move onto the next topic, rather, acknowledge the fact that the person is engaging and partaking in the sessions. Most of the activities are designed to be 'open' with several possible answers.

3

Putting the key principles into practice

4. Having a tangible focus – something to look at or feel.

Multi-sensory cues are really important, as memory works much better if you do not rely on just one sense. Try to have a mix of activities involving vision, touch, hearing, taste and smell. Often it is a combination of senses that is most effective.

- Having something to look at or touch really helps to aid concentration. Words in a discussion may soon be lost when memory is limited; having an object, a photograph or picture keeps the person's attention on the activity.

5. Using Reminiscence

Using past memories is an excellent way of tapping into a strength that many people with memory problems have, in terms of recalling experiences from much earlier in their lives. The manual contains many activities that allow you to compare old and new, and think about how things have changed over time. Remember though that some people may have unhappy memories of their earlier life, and some sensitivity is needed.

6. Maximising Potential

Be careful not to assume the person is unable to contribute or carry out an activity simply because they were not able to yesterday or last week. People with memory problems often function at less than their full potential, perhaps due to lack of stimulation or opportunity.

Tips

- Keep an **open mind** when choosing activities
- Give the person **time** to gather their thoughts or carry out an activity
- **Do not overload or overwhelm** them with information
- Provide just enough **prompting** to enable the person to do the activities themselves

4

Putting the key principles into practice

7. Enjoyable and fun

A key goal of the therapy is that the activities make your partner feel enabled and empowered. If you find your partner commenting that "This is like being back in school", something is going wrong! The activities should provide a learning atmosphere which is fun and enjoyable.

Tip

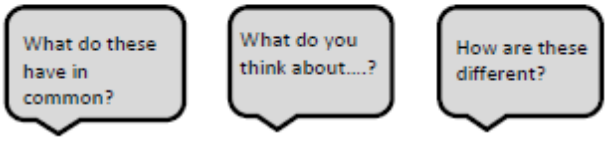
If your partner makes comments about school, ask them what they liked and disliked about school. Reflect on whether you are taking on the role of 'teacher' too readily.

8. Stimulating new ideas and communication

Often with people with memory problems, we tend to talk about things from the past. Whilst this is enjoyable for people, it often involves recalling information, that has been over-rehearsed. The aim of the this therapy is to continually encourage new ideas, thoughts and associations, rather than just recall previously learned information.

Problem solving: How do I encourage discussion?

- By asking questions: The way you phrase questions is important in encouraging the person to explore ideas. Try these examples:



- By introducing a variety of topics: Encourage discussions about new topics that you haven't talked about before. Use the examples provided in the manual to get you started.

9. Strengthening the caregiving relationship

The activities present a great opportunity for you and your partner to enjoy some quality time together and it is important that both of you feel relaxed and calm before starting with the session. Get plenty of rest and complete the topic at your own pace and time or alternatively, pick another date and time to complete the topic.

5

Role playing exercise

The purpose of these exercises is to demonstrate some examples of good and bad practice in iCST. The exercises give the caregiver the opportunity to experience what it might be like to take part in iCST from the perspective of someone living with dementia.

ROLES: Researcher = 'caregiver'; caregiver being trained – person with dementia

Bad practice example 1 (fact based questions and putting the Person with dementia 'on the spot')

Carer will ask the person with dementia some closed, factual questions such as: "what is the date today" and will prompt with a comment such as " come on, you know this".

Bad practice example 2 (overloading the Person with dementia, adding pressure, taking a teacher-like role)

Carer explains the activity to the Person with dementia very quickly and offers lots of information e.g: "now we are going to look at some photographs and I want you to tell me where they were taken and what year you think they were taken. Can you also give me some other examples of important landmarks in Blackpool?

Bad practice example 3 (correcting the person, focusing on weakness rather than strength)

Carer asks the Person with dementia to tell them what are the main differences between the photos (number of people, beach vs pub). Even if they get the answer right, the carer comments "No, that's not the main difference, come on, can't you spot it?"

……

Good practice example 1 (person centred approach, making the therapy 'their own')

Carer offers the Person with dementia the opportunity to choose which activity they would like to do (Blackpool or Moon landing)

Good practice example 2 (encouraging discussion with open questions, focus on opinions not facts)

Carer shows the Person with dementia the chosen topic and asks "which of these images do you like the most?" and "What might you do if you went to the seaside/ what would you do if you went into space?"

Good practice example 3 (completing activities together, having a tangible focus e.g. images)

Carer as the Person with dementia " can you see any similarities between the photos? Is this a place you would like to visit? Why/why not? Would there be any difference in Summer or Winter? What would the differences be.....Carer will be very positive and give praise when tasks are complete.

INVEST Companion diary

How was today's session?

Topic title: _____ Page: _____ Date:_____

Did you finish today's topic? YES NO

If no, please state reason. _____

Did you partially complete today's topic then switch to another topic partway? YES NO
If so, to which one (*Title, page*): _____

How much time did you spend on today's topic (in minutes)? _____

Please select the most appropriate response to the statements below:

During the session....	Strongly Disagree	Disagree	Neutral	Agree	Strong Agree
your partner was interested.	1	2	3	4	5
your partner was motivated	1	2	3	4	5
your partner displayed emotional responses*	1	2	3	4	5
your partner took the initiative (at times)	1	2	3	4	5
your partner gained a sense of achievement	1	2	3	4	5

(*anger, disgust, fear, happiness, surprise, sadness)

*About **you**.....*

you felt confident using iCST	1	2	3	4	5

What did you like most about the session today?

_____

What did you like least about the session today?

_____

Through your support, what aspect of the session did your partner find the most fulfilling?

_____

Can you offer any comments and ideas to improve the session?

_____

Appendix F: Postal questionnaire documents

Participant Invitation Letter

A questionnaire survey for spouses/partners of people with Parkinson's-related dementias

Dear .....,

You are invited to take part in a study to better understand the experiences of spouses and life partners of people with Parkinson's disease and cognitive impairment. The study focuses on exploring personal experiences and feelings of life partners and spouses who are providing care and support to their loved ones with either Parkinson's disease and mild cognitive impairment, Parkinson's disease dementia or Dementia with Lewy bodies. Participation in this study would require you to complete the questionnaire which is posted together with this letter.

Attached is a participant information sheet providing further details of the research study. Please take some take to read this document before deciding whether or not you are interested in taking part. Taking part in the study is voluntary. If you do decide to take part please complete the consent form and questionnaire attached to this letter and post both documents back to the research team in the prepaid envelope. If you do not wish to take part, please dispose of the questionnaire.

If you have any questions please do not hesitate to contact:

- Ms Sabina Vatter on 0161 306 7913 or sabina.vatter@manchester.ac.uk

Thank you for considering taking part in our research, I look forward to talking to you soon.

Best wishes,

Sabina, on behalf of Dr Iracema Leroi (0161 306 7913)

PARTICIPANT INFORMATION SHEET

A questionnaire survey for spouses/partners of people with Parkinson's-related dementias

Invitation to participate in a research study

We would like to invite you to take part in a research study that involves filling in a short paper-based questionnaire to explore the experiences of spouses and life partners of people with Parkinson's disease and cognitive impairment. This study is part of a PhD project being used for educational purposes.

This information sheet explains what taking part in this study would involve. Before you decide, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Thank you for reading this information sheet.

What is the purpose of the study?

This is a paper-based questionnaire study aimed at exploring personal experiences and feelings of life partners and spouses who are providing care and support to their loved ones with either Parkinson's disease and mild cognitive impairment, Parkinson's disease dementia or Dementia with Lewy bodies. The survey includes questions about own health and well-being, resilience, associated feelings of providing care as well as questions regarding relationship satisfaction.

Why have I been invited?

You have been invited because you care for a partner who has Parkinson's-related cognitive impairment.

What will I be required to do?

We will ask you to complete a paper-based questionnaire about your health and personal experiences of providing care to your partner or spouse. The questionnaire will take approximately 15 – 20 minutes to complete. You will be provided with a pre-paid envelope for you to use to return the completed questionnaire and the signed and dated consent form. This information sheet is for you to keep. Alternatively, the questionnaire can be filled in the presence of one of our researchers who can assist you should you wish to do so.

Can I choose whether or not to take part?
Yes. It is up to you to decide whether or not you would like to take part. If you do wish to take part, you are asked to sign a consent form that will allow us to use the information you provide and to complete the questionnaire. If you do not wish to take part, then please dispose of the questionnaire.

How will this study benefit me?
Taking part in this study may not directly benefit you however your involvement will contribute significantly to our wider understanding of the experiences of life partners and spouses. It will increase knowledge in the scientific community and improve our understanding of how we can best support spouses and life partners.

Will my GP be informed about my involvement in this study?
Your GP will not be notified about your participation in this study.

Will my participation in the study be kept confidential?
Your participation and all the information we collect about you will be kept confidential. All the information gathered about you for the questionnaire will be completely anonymous and cannot be linked to your personal information.

Only the study team at the University of Manchester will have access to your personal information. Data from the questionnaire surveys will be held securely in a separate filing system to your personal information so that you cannot be identified from your responses and will be carefully destroyed as soon as it is not needed. The anonymous research data will be stored at the University of Manchester. The data will be analysed by researchers involved in this study.

Individuals from the University, regulatory authorities or NHS Trust may need to access the data collected to ensure that the study is being carried out properly. This is to protect you by ensuring that we are doing the research in a safe and ethical way. All individuals will be authorised representatives from each organisation and will have a duty of confidentiality to all research participants.

It is also possible that the data collected in this study might be useful for other research taking place in the future. If we used your data in future research, it would all be anonymous for both the analysis and reporting of results.

If you have any questions about what will happen to your data during the study please contact the research team whose details are at the end of this document.

What will happen to the results of the study?
When the study is complete, we will present the findings at conferences and publish them in scientific journals. All your data will be anonymised and you will not be identified personally. If you would like a short summary at the end of the study please let the research team know. Upon completion, a summary can be posted at your home address.

What if there is a problem?
If you have a concern about any aspect of this study, please ask to speak to the researchers who will do their best to answer your questions.

If there are any issues regarding this research that you would prefer not to discuss with members of the research team, please contact the Research Governance and Integrity Team by either writing to 'The Research Governance and Integrity Manager, Research Office, Christie Building, The University of Manchester, Oxford Road, Manchester M13 9PL', by emailing: Research.Complaints@manchester.ac.uk, or by telephoning 0161 275 7583 or 275 8093.

In the event that something does go wrong and you are harmed during the research you may have grounds for a legal action for compensation against the University of Manchester but you may have to pay your legal costs.

Who has reviewed the study?
This research has been looked at by an independent group of people, called a Research Ethics Committee to protect your safety, rights, well-being and dignity. This study has been reviewed and been given a favourable opinion by the NRES Yorkshire & The Humber-Bradford Leeds Ethics Committee.

Who can I contact for further information?
If you would like further information or have any questions about the study, please contact the following researcher:

Ms Sabina Vatter (Research Assistant and PhD student)
University of Manchester,
Division of Neuroscience and Experimental Psychology,
Jean McFarlane Building, 3rd floor,
Oxford Road, Manchester, M13 9PL
Tel: 0161 306 7913
Email: sabina.vatter@manchester.ac.uk

Thank you for considering taking part in this research study.

CONFIDENTIAL

Project title:
A questionnaire survey for spouses/partners of people with Parkinson's-related dementias

Chief Investigator: Dr. Iracema Leroi

	Please initial box to confirm consent
1. I confirm that I have read and understood the Participant Information Sheet dated 19th of May 2017 (version 1.0) for the above study and have had the opportunity to ask questions.	Initial
2. I understand that my participation in this study is voluntary and that I am free to withdraw at any time, without giving any reason, without my medical care and legal rights being affected.	Initial
3. I understand that all information given by me or about me will be treated as confidential by the research team and that I will not be identified in any way in the analysis and reporting of the results.	Initial
4. I understand that the information collected about me during this study may be used to support future research and may be shared anonymously with the other researchers.	Initial
5. I understand that data collected during the study may be looked at by individuals from the University of Manchester, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to the data.	Initial
6. I agree to be contacted about future studies: (Optional) Yes or No: ……	Initial
7. I agree to take part in the above study.	Initial

Name of participant	Date	Signature
Name of researcher	Date	Signature

Participant ID ☐

Questionnaire Survey

Thank you for agreeing to participate in this study. In this booklet you will find 9 short questionnaires. Please read the general instructions below before completing the questionnaires, there are no right or wrong answers.

Should you have any difficulties, please contact the researcher for assistance:

- Ms Sabina Vatter on 0161 306 7913 or sabina.vatter@manchester.ac.uk

General instructions

To ensure the smooth operation of the data management, it would be appreciated if the following could be observed:

- Please complete the form using a black or blue pen
- Please do not crease the form, you may fold it once
- Please complete all the questions
- Please enter your responses in the boxes/ spaces provided, as instructed
- Please use only a single line to delete mistakes and initial each such correction

Thank you for completing the questionnaire!

Today's date (dd/mm/yyyy): ☐☐☐☐☐☐☐☐

Has your partner been diagnosed with any of the following? If so, please tick:

- ☐ Parkinson's disease and mild cognitive impairment
- ☐ Parkinson's disease dementia
- ☐ Dementia with Lewy Bodies
- ☐ Other (please specify): .....

Which clinic or hospital does your partner normally attend? Please tick:

- ☐ Greater Manchester Mental Health NHS Foundation Trust
- ☐ Salford Royal Foundation Trust
- ☐ Pennine Acute Trust
- ☐ University Hospital of South Manchester NHS Foundation Trust
- ☐ North West Borough Healthcare NHS Foundation Trust
- ☐ Other (please specify): .....

Participant ID ☐

Sociodemographic Questionnaire: Background details

To help with our study it will be helpful to have some background details about you and your partner. These will allow us to compare groups in the study with the general population. All data is confidential and stored in an anonymised form.

1. What is your relationship to the participant?
*Please tick **one** box only*

a. Wife/Husband (spouse) ☐
b. Partner ☐
c. Other (please specify) ☐ .....

2. Please indicate the living status of yourself and your partner
*Please tick **all** that applies*

You live with:	Spouse/partner ☐	Other family ☐	Other ☐
Partner lives with:	Spouse/partner ☐	Other family ☐	Other ☐

3. How long have you known each other? ..... years

4. How long have you been in a relationship / married? ..... years

5. How long have you been caring for your partner (if applicable)? ..... years

6. Approximately how many hours a day do you spend on caring? ..... hours

7. Please indicate your and your partner's gender
Please tick as appropriate

Your gender	Male ☐	Female ☐
Partner's gender	Male ☐	Female ☐

8. Please indicate your and your partner's date of birth

Your DOB (dd/mm/yyyy): ☐☐ ☐☐ ☐☐☐☐

Partner's DOB (dd/mm/yyyy): ☐☐ ☐☐ ☐☐☐☐

9. Please indicate the age you and your partner left full-time education?

You ☐☐ Not applicable (i.e. never had formal education) ☐

Partner ☐☐ Not applicable (i.e. never had formal education) ☐

10. Please indicate the highest level of education of yourself and your partner:
*Please tick **one** box only*

You	Partner
☐ School Leaver (14-16 years of age)	☐ School Leaver (14-16 years of age)
☐ School Leaver (18 years of age)	☐ School Leaver (18 years of age)
☐ Further Education (Vocational qualifications: i.e. GNVQ/NVQ/HND)	☐ Further Education (Vocational qualifications: i.e. GNVQ/NVQ/HND)
☐ Higher Education (BSc/BA or equivalent)	☐ Higher Education (BSc/BA or equivalent)
☐ Postgraduate Education (MSc/MA/PhD or equivalent)	☐ Postgraduate Education (MSc/MA/PhD or equivalent)

11. Please indicate your and your partner's ethnicity
*Please tick **one** box only for each*

You

WHITE	☐ White British ☐ White Irish ☐ Other White background
BLACK or BLACK BRITISH	☐ Caribbean ☐ African ☐ Other Black background
MIXED	☐ White & Black Caribbean ☐ White & Black African ☐ White & Asian ☐ Other Mixed Background
ASIAN or ASIAN BRITISH	☐ Indian ☐ Pakistani ☐ Bangladeshi ☐ Other Asian background
CHINESE	☐ Chinese
OTHER	☐ Other ethnic group ☐ Not stated ☐ Do not wish to specify

Your partner

WHITE	☐ White British ☐ White Irish ☐ Other White background
BLACK or BLACK BRITISH	☐ Caribbean ☐ African ☐ Other Black background
MIXED	☐ White & Black Caribbean ☐ White & Black African ☐ White & Asian ☐ Other Mixed Background
ASIAN or ASIAN BRITISH	☐ Indian ☐ Pakistani ☐ Bangladeshi ☐ Other Asian background
CHINESE	☐ Chinese
OTHER	☐ Other ethnic group ☐ Not stated ☐ Do not wish to specify

12. Please answer the following questions about your partner:

- What year was your partner diagnosed with Parkinson's disease or Dementia with Lewy Bodies? ☐☐☐☐
- What year did you notice any symptoms of cognitive impairment? ☐☐☐☐

Participant ID ☐

Questionnaire 1: EuroQol EQ-5D

By placing a tick in one box in each group below, please indicate which statements best describe your own health state today.

Mobility

I have no problems in walking about ☐
I have some problems in walking about ☐
I am confined to bed ☐

Self-care

I have no problems with self-care ☐
I have some problems with washing or dressing myself ☐
I am unable to wash or dress myself ☐

Usual activities (*e.g. work, study, housework, family or leisure activities*)

I have no problems with performing my usual activities ☐
I have some problems with performing my usual activities ☐
I am unable to perform my usual activities ☐

Pain/Discomfort

I have no pain or discomfort ☐
I have moderate pain or discomfort ☐
I have extreme pain or discomfort ☐

Anxiety/Depression

I am not anxious or depressed ☐
I am moderately anxious or depressed ☐
I am extremely anxious or depressed ☐

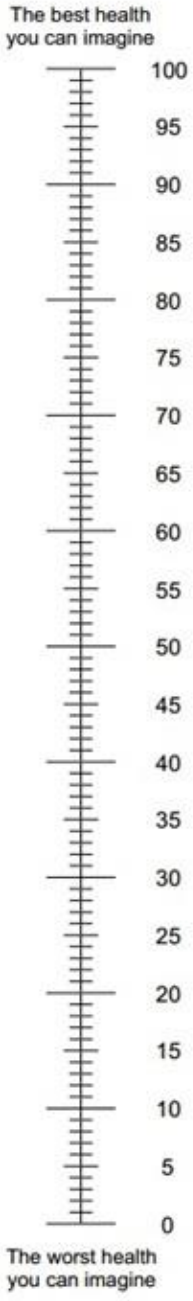
Participant ID

EuroQol EQ-5D

To help people say how good or bad a health state is, we have drawn a scale (rather like a thermometer) on which the best state you can imagine is marked 100 and the worst state you can imagine is marked 0.

We would like you to indicate on this scale how good or bad your own health is TODAY, in your opinion. Please do this by drawing a line from the box below to whichever point on the scale indicating how good or bad your health state is today.

Your health state today



Participant ID

Questionnaire 2: Relationship Satisfaction Scale

Please circle the number on each row that best describes the amount of satisfaction you feel with your partner.

	Very dissatisfied	Moderately dissatisfied	Somewhat dissatisfied	Neutral	Somewhat satisfied	Moderately satisfied	Very satisfied
1. Communication and openness	0	1	2	3	4	5	6
2. Resolving conflicts and arguments	0	1	2	3	4	5	6
3. Degree of affection and caring	0	1	2	3	4	5	6
4. Intimacy and closeness	0	1	2	3	4	5	6
5. Satisfaction with your role in the relationship	0	1	2	3	4	5	6
6. Satisfaction with the other person's role in the relationship	0	1	2	3	4	5	6
7. Overall satisfaction with the relationship	0	1	2	3	4	5	6

Participant ID

Questionnaire 3: Brief Resilience Scale

Please respond to each item by circling the number on each row.

	Strongly disagree	Disagree	Neutral	Agree	Strongly agree
1. I tend to bounce back quickly after hard times.	1	2	3	4	5
2. I have a hard time making it through stressful events.	5	4	3	2	1
3. It does not take me long to recover from a stressful event.	1	2	3	4	5
4. It is hard for me to snap back when something bad happens.	5	4	3	2	1
5. I usually come through difficult times with little trouble.	1	2	3	4	5
6. I tend to take a long time to get over set-backs in my life.	5	4	3	2	1

Questionnaire continues on the next page...

Participant ID

Questionnaire 4: SF-12® Health Survey

This survey asks for your views about your health. This information will help you keep track of how you feel and how well you are able to do your usual activities.
Answer every question by selecting the answer as indicated. If you are unsure about how to answer a question, please give the best answer you can.

1. In general, would you say your health is:

Excellent	Very good	Good	Fair	Poor
○	○	○	○	○

2. The following questions are about activities you might do during a typical day. Does your health now limit you in these activities? If so, how much?

	Yes, limited a lot	Yes, limited a little	No, not limited at all
a. Moderate activities, such as moving a table, pushing a vacuum cleaner, bowling, or playing golf			
b. Climbing several flights of stairs			

3. During the past 4 weeks, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of your physical health?

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a. Accomplished less than you would like					
b. Were limited in the kind of work or other activities					

4. During the past 4 weeks, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)?

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a. Accomplished less than you would like					
b. Did work or other activities less carefully than usual					

5. During the past 4 weeks, how much did pain interfere with your normal work (including both work outside the home and housework)?

Not at all	A little bit	Moderately	Quite a bit	Extremely
○	○	○	○	○

Questionnaire 4 continues…

6. **These questions are about how you feel and how things have been with you during the past 4 weeks. For each question, please give the one answer that comes closest to the way you have been feeling.**

How much of the time during the past 4 weeks…

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a. Have you felt calm and peaceful?					
b. Did you have a lot of energy?					
d. Have you felt downhearted and blue?					

7. **During the past 4 weeks, how much of the time has your physical health or emotional problems interfered with your social activities (like visiting friends, relatives, etc.)?**

All of the time ○ Most of the time ○ Some of the time ○ A little of the time ○ None of the time ○

Questionnaire continues on the next page…

Participant ID

Questionnaire 5: Hospital Anxiety and Depression Scale

Instructions: This questionnaire asks about how you feel. Read each item and circle the reply which comes closest to how you have been feeling in the past week. Don't take too long over your replies: your immediate reaction to each item will probably be more accurate than a long thought out response.

1. I feel tense or 'wound up':	**A**
Most of the time	3
A lot of the time	2
Time to time, occasionally	1
Not at all	0

2. I still enjoy the things I used to enjoy:	**D**
Definitely as much	0
Not quite so much	1
Only a little	2
Not at all	3

3. I get a sort of frightened feeling like something awful is about to happen:	**A**
Very definitely and quite badly	3
Yes, but not too badly	2
A little, but it doesn't worry me	1
Not at all	0

4. I can laugh and see the funny side of things:	**D**
As much as I always could	0
Not quite so much now	1
Definitely not so much now	2
Not at all	3

5. Worrying thoughts go through my mind:	**A**
A great deal of the time	3
A lot of the time	2
From time to time but not too often	1
Only occasionally	0

6. I feel cheerful:	**D**
Not at all	3
Not often	2
Sometimes	1
Most of the time	0

7. I can sit at ease and feel relaxed:	**A**
Definitely	0
Usually	1
Not often	2
Not at all	3

8. I feel as if I am slowed down:	**D**
Nearly all of the time	3
Very often	2
Sometimes	1
Not at all	0

9. I get a sort of frightened feeling like 'butterflies in the stomach':	**A**
Not at all	0
Occasionally	1
Quite often	2
Very often	3

10. I have lost interest in my appearance:	**D**
Definitely	3
I don't take as much care as I should	2
I may not take quite as much care	1
I take just as much care as ever	0

11. I feel restless as if I have to be on the move:	**A**
Very much indeed	3
Quite a lot	2
Not very much	1
Not at all	0

12. I look forward with enjoyment to things:	**D**
A much as I ever did	0
Rather less than I used to	1
Definitely less than I used to	2
Hardly at all	3

13. I get sudden feelings of panic:	**A**
Very often indeed	3
Quite often	2
Not very often	1
Not at all	0

14. I can enjoy a good book or radio or TV programme:	**D**
Often	0
Sometimes	1
Not often	2
Very seldom	3

Participant ID

Questionnaire 6: Zarit Burden Interview.

The following is a list of statements which reflect how people sometimes feel when taking care of another person. After each statement, indicate how often you feel that way: never, rarely, sometimes, quite frequently, or nearly always. There are no right or wrong answers.

	Never	Rarely	Some-times	Quite frequently	Nearly always
1. Do you feel that your partner asks for more help than he or she needs?	0	1	2	3	4
2. Do you feel that, because of the time you spend with your partner, you don't have enough time for yourself?	0	1	2	3	4
3. Do you feel stressed between caring for your partner and trying to meet other responsibilities for your family or work?	0	1	2	3	4
4. Do you feel embarrassed about your partner's behaviour?	0	1	2	3	4
5. Do you feel angry when you are around your partner?	0	1	2	3	4
6. Do you feel that your partner currently affects your relationship with other family members?	0	1	2	3	4
7. Are you afraid about what the future holds for your partner?	0	1	2	3	4
8. Do you feel that your partner is dependent upon you?	0	1	2	3	4
9. Do you feel strained when you are around your partner?	0	1	2	3	4
10. Do you feel that your health has suffered because of your involvement with your partner?	0	1	2	3	4
11. Do you feel that you don't have as much privacy as you would like, because of your partner?	0	1	2	3	4
12. Do you feel that your social life has suffered because you are caring for your partner?	0	1	2	3	4
13. Do you feel uncomfortable having your friends over because of your partner?	0	1	2	3	4
14. Do you feel that your partner seems to expect you to take care of him or her, as if you were the only one he or she could depend on?	0	1	2	3	4
15. Do you feel that you don't have enough money to care for your partner, in addition to the rest of your expenses?	0	1	2	3	4
16. Do you feel that you will be unable to take care of your partner much longer?	0	1	2	3	4
17. Do you feel that you have lost control of your life since your partner's illness?	0	1	2	3	4
18. Do you wish that you could just leave the care of your partner to someone else?	0	1	2	3	4
19. Do you feel uncertain about what to do about your partner?	0	1	2	3	4
20. Do you feel that you should be doing more for your partner?	0	1	2	3	4
21. Do you feel that you could do a better job in caring for your partner?	0	1	2	3	4
22. Overall, how burdened do you feel in caring for your partner?	0	1	2	3	4

Participant ID

Questionnaire 7: Dyadic Relationship Scale

Please respond to each item by circling the number on each row.

	Strongly disagree	Disagree	Agree	Strongly agree
1. I felt closer to her/him than I have in awhile.	0	1	2	3
2. I have learned some good things about my (partner).	0	1	2	3
3. I felt angry toward him/her.	3	2	1	0
4. I felt depressed because of my relationship with him/her.	3	2	1	0
5. I felt resentful toward him/her.	3	2	1	0
6. I have had more patience than I have had in the past.	0	1	2	3
7. I have learned some good things about myself.	0	1	2	3
8. I felt that my relationship with him/her was strained.	3	2	1	0
9. I have learned some nice things about other people in my life.	0	1	2	3
10. Communication between my (partner) and me has improved.	0	1	2	3
11. I felt that he/she made requests over and above what he/she needed.	3	2	1	0

Questionnaire continues on the next page…

Questionnaire 8: Relatives' Stress Scale

Please respond to each item by circling the number on each row.

	Never/ not at all	Rarely/ a little	Sometimes/ moderately	Frequently/ quite a lot	Always/ considerably
1. Do you ever feel you can no longer cope with the situation?	0	1	2	3	4
2. Do you ever feel that you need a break?	0	1	2	3	4
3. Do you ever get depressed by the situation?	0	1	2	3	4
4. Has your own health suffered at all?	0	1	2	3	4
5. Do you worry about accident happening to (*your partner)*?	0	1	2	3	4
6. Do you ever feel that there will be no end to the problem?	0	1	2	3	4
7. Do you find it difficult to get away on holiday?	0	1	2	3	4
8. How much has your social life been affected?	0	1	2	3	4
9. How much has the household routine been upset?	0	1	2	3	4
10. Is your sleep interrupted by *(your partner)*?	0	1	2	3	4
11. Has your standard of living been reduced?	0	1	2	3	4
12. Do you ever feel embarrassed by (*your partner)*?	0	1	2	3	4
13. Are you at all prevented from having visitors?	0	1	2	3	4
14. Do you ever get cross and angry with *(your partner)*?	0	1	2	3	4
15. Do you ever feel frustrated at times with *(your partner)*?	0	1	2	3	4

Participant ID

Questionnaire 9: Family caregiving role

Please respond to each item by circling the number on each row.

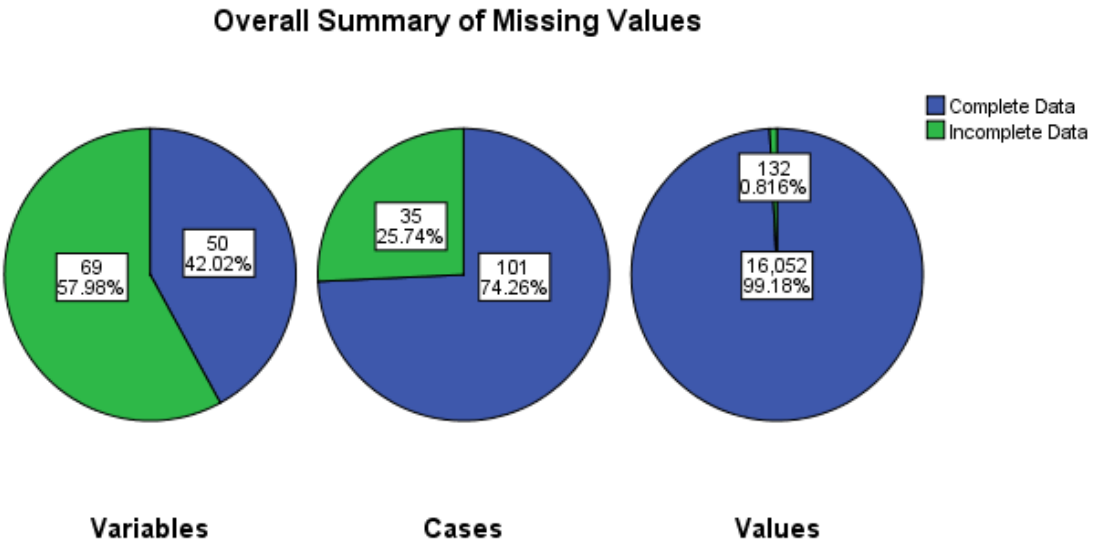
	Strongly disagree	Disagree	Not sure	Agree	Strongly agree
1. I get satisfaction seeing (name) accomplish things.	1	2	3	4	5
2. Caring for (name) has made me more confident dealing with others.	1	2	3	4	5
3. I get a great deal of satisfaction from caring.	1	2	3	4	5
4. I would really feel at a loss if (name) was not around.	1	2	3	4	5
5. I worry about what would happen to (name) if something happened to me.	1	2	3	4	5
6. I feel reassured knowing that as long as I am helping (name) he/she is getting proper care.	1	2	3	4	5
7. I grieve for the opportunities (name) doesn't have.	1	2	3	4	5
8. I have lost control of my life since caring for (name).	1	2	3	4	5
9. I regret the opportunities I don't have.	1	2	3	4	5
10. My friends don't visit as often because I am caring for (name).	1	2	3	4	5
11. I care for (name) because no-one else can.	1	2	3	4	5
12. Caring for (name) takes up most of my time and I neglect the rest of the family.	1	2	3	4	5
13. I am angry when I am around (name).	1	2	3	4	5
14. Nothing I can do seems to please (name).	1	2	3	4	5
15. I am embarrassed over (name's) behaviour.	1	2	3	4	5
16. I feel guilty about (name).	1	2	3	4	5

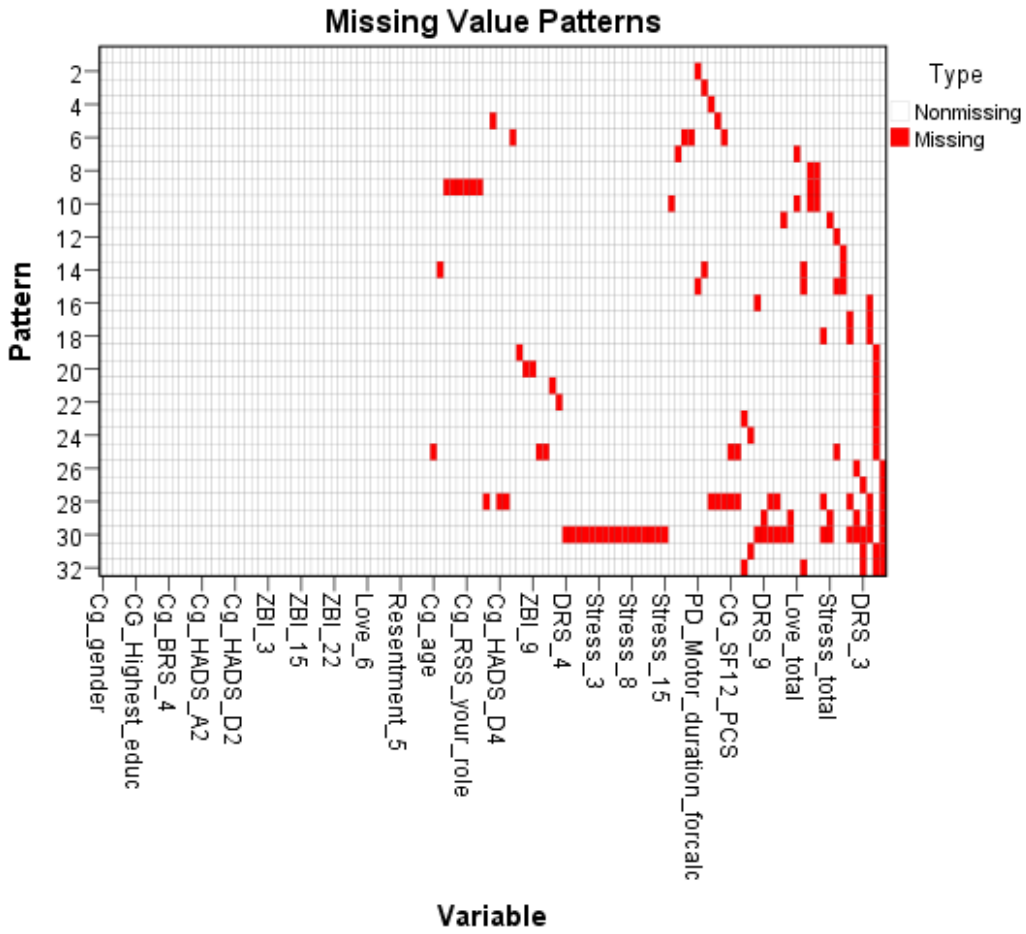
Thank you for completing the questionnaire!

Your help is much appreciated!

Appendix G: Study 1 documents (Chapter 5)

Missing values in Study 1





Assumption tests in Study 1

In order to perform Pearson correlation analyses, the variables should be continuous, have normal distribution, linearity, homoscedasticity and no outliers. The variables in the current study are continuous, which meets the first assumption. The second assumption, which denotes that variables should be normally distributed, was not met for the majority of values according to the Shapiro-Wilk's test and visual inspection of histograms. However, none of the values were extremely skewed (i.e. within ±1.96), and only one variable (the EQ-5D index score) appeared to be kurtotic (= 3.824). The assumption of linearity was met for all variables according to the scatterplot matrix. According to the residual plots, all variables had homoscedasticity; therefore, the fourth assumption was met. The fifth assumption looked at whether there were any outliers in the data. A visual inspection of boxplots revealed that three variables (EQ-5D index, EQ-5D-VAS and FCR-satisfaction sub-scale) had outliers. These outliers were looked through in the dataset to verify that they were not due to data entry errors; rather, they were low values compared to the rest of the sample. In order to observe whether the outliers affected the output, some analyses were run with outliers and some with transformed outliers, as appropriate. The outliers were transformed using winsorization, whereby outliers were assigned the highest or the lowest value found in the sample which was not an outlier.

Since not all the assumptions were met for the Pearson correlation analyses, it was decided that a non-parametric (i.e. Spearman correlation) would be used instead. Spearman correlation analyses met the required assumptions for conducting a correlation analyses, including that the variables should be measured on an ordinal or interval scale (i.e. continuous) and there is a monotonic pattern between the variables, as observed in the scatterplots. Spearman correlation is also not very sensitive to outliers; thus, the analysis was performed with the outliers included.

The required assumptions for an ANOVA and independent t-test included: (1) continuous variables, which was met; (2) presence of at least two independent groups, which was met; (3) independence of observations, which was met as no participants were in more than one group simultaneously; (4) approximately normally distributed data, which was not met for the majority of variables; however, ANOVA is considered relatively robust to some violation of normality, (5) no outliers, which were identified and winsorized; and (6) homogeneity of variances (i.e. Levene's test $p > 0.05$), which was met for all variables except HADS-depression, FCR-satisfaction and care provision years, and a Welch F test for these three variables was used instead. For the variables that met the assumption, ANOVA and t-tests were used. For the variables that failed to meet these assumptions, the corresponding non-parametric tests were performed (i.e. Kruskal-Wallis H test and the Mann-Whitney U-test).

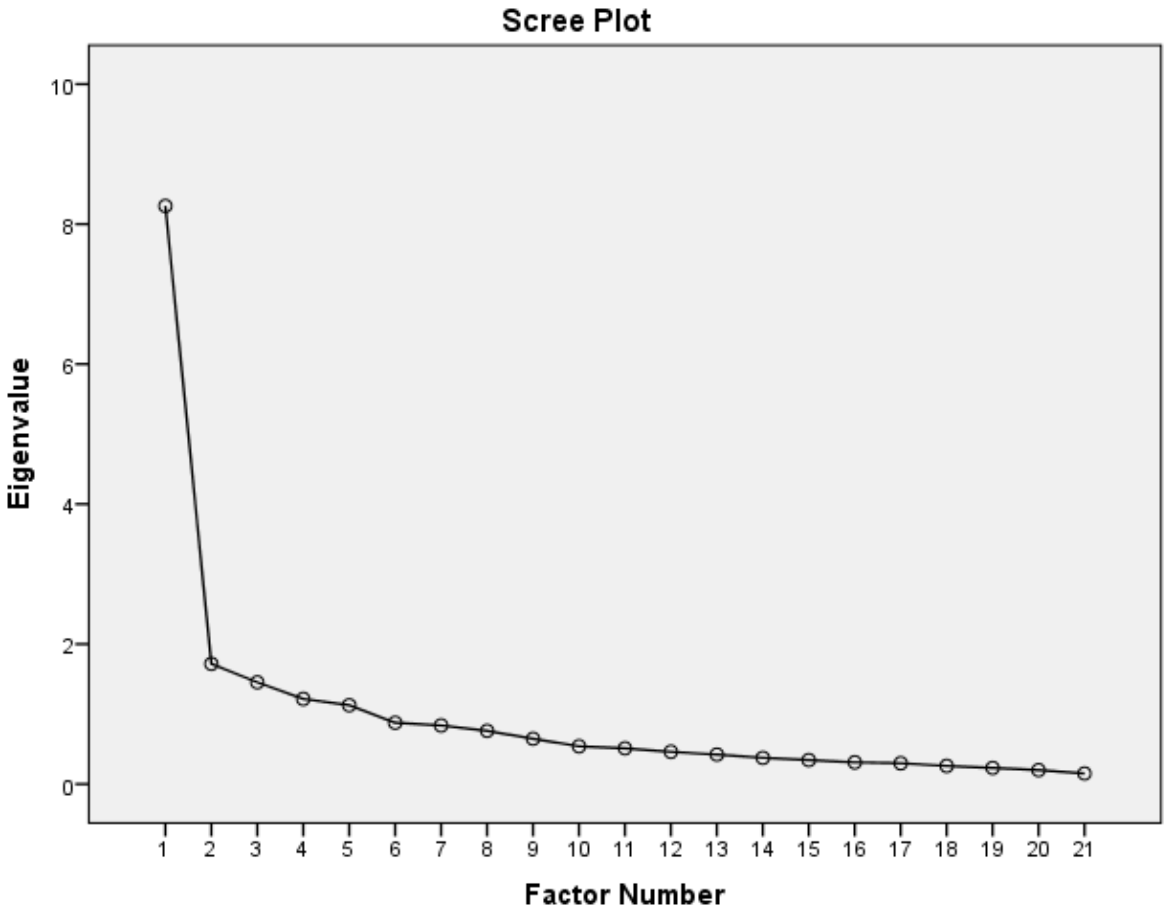
Table G.1 Assumption tests of parametric tests for each outcome variable.

Measures Assumptions	RSS	ZBI	BRS	HADS -A	HADS -D	SF-12 PCS	SF-12 MCS	EQ5D index	EQ5D VAS	Rel. SS	DRS-inter	DRS-strain	FCR satisf	FCR resent	FCR anger	Care-giving years	Care-giving h/pw
1. Continuous variable (i.e. interval, ratio)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
2. Normal distribution																	
(a) Shapiro-Wilk's test (p > 0.05)	(a) **X** 0.000	(a) ✓ 0.499	(a) **X** 0.013	(a) **X** 0.002	(a) **X** 0.000	(a) **X** 0.000	(a) **X** 0.004	(a) **X** 0.000	(a) **X** 0.000	(a) ✓ 0.063	(a) ✓ 0.193	(a) **X** 0.000	(a) **X** 0.001	(a) **X** 0.038	(a) **X** 0.000	(a) **X** 0.000	(a) **X** 0.000
(b) histograms	(b) **X**	(b) ✓	(b) ✓	(b) **X**	(b) **X**	(b) **X**	(b) **X**	(b) **X**	(b) **X**	(b) ✓	(b) ✓	(b) **X**	(b) **X**	(b) ✓	(b) **X**	(b) **X**	(b) **X**
3. Linearity (scatterplot matrix)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
4. Homoscedasticity (residual plots)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
5. Outliers: (boxplots)	✓	✓	✓	✓	✓	✓	✓	9	1	✓	✓	✓	2	✓	✓	10	✓
6. Independence of observations	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
7. Homogeneity of variance (Levene's test, p > 0.05)	0.448 ✓	0.205 ✓	0.514 ✓	0.135 ✓	0.001 **X** Welch (14.945), p= 0.000	0.479 ✓	0.446 ✓	0.261 ✓	0.137 ✓	0.740 ✓	0.229 ✓	0.531 ✓	0.002 **X** Welch (0.318), p = 0.729	0.170 ✓	0.496 ✓	0.000 **X** Welch (9.364), p= 0.000	0.521 ✓

Abbreviations: BRS – Brief Resilience Scale; DRS – Dyadic Relationship Scale, positive interaction or negative strain sub-scale; EQ-5D – EuroQoL-5D index or visual analogue scale (VAS); FCR – Family Caregiving Role scale, caregiving satisfaction, resentment or anger sub-scale; HADS – Hospital Anxiety and Depression Scale, anxiety or depression sub-scale; Rel.SS – Relatives' Stress Scale; RSS – Relationship Satisfaction Scale; SF-12 – Short Form 12 Health Survey, physical health (PCS) or mental health (MCS) sub-scale; ZBI – Zarit Burden Interview.

Appendix H: Study 2 documents (Chapter 6)

The scree plot of the exploratory factor analysis:



Methodological description of the assumption tests:

Prior to conducting multiple linear regression analyses, the following assumption tests were carried out:

(1) Appropriate sample size: Is the sample above 104 + m (number of predictors)?
(2) Linearity between independent and dependent variables: Is the relationship between the dependent variable(s) and independent variables linear? (observed in the matrix of scatterplots)
(3) Independence of residuals: Are the residuals independent? [assessed with Durbin-Watson test; the acceptable range for the Durbin-Watson test is provided according to the sample size and number of predictors (Durbin & Watson, 1951; Field, 2013)]
(4) Lack of multicollinearity: Is there a medium to weak ($r \leq 0.8$) correlation between the independent variables? A high correlation between independent variables indicates collinearity and the variables with strong correlations should be re-evaluated and removed. [collinearity is observed with the correlation matrix; tolerance (acceptable range between 0.2 – 0.9); variance inflation factor, VIF (acceptable values below 10)]
(5) Homoscedasticity: Is the variation in the residuals constant? (assessed with residual plots)
(6) Distribution of residuals: Are the residuals distributed normally? [observed in the probability-probability plot (P-P plot)]
(7) Outliers: Are there any extreme values (outliers) in the data? [assessed with Cook's distance (values above 1 were removed), and with Mahalanobis distance (acceptable range depended on the degree of freedom (= the number of predictors) and a chi square distribution (Field, 2013)].

Assumption tests for multiple linear regression analyses:

The sample size was appropriate for each factor (104 participants + 7 predictors = 111). There was a linear relationship between the dependent variables (factors 1 to 5) and the independent variables (predictors) as observed in the matrix of scatterplots on all five factors (see Appendix H). According to the Durbin-Watson statistic the values of residuals on factor 1 were independent, where with seven predictors and 117 cases at the 5% significance level a range between 1.592 and 1.807 was allowed (Stanford University). The Durbin-Watson statistic for factors 2 – 5, which varied between 1.838 and 2.112 was also considered acceptable as a value close to 2 indicated that the residuals were uncorrelated (Field, 2013); therefore, the assumption that the residuals were independent was met. Analysis of collinearity statistics was evaluated with correlation matrix, tolerance level (between 0.27 and 0.49 across all factors) and variance inflation factor (VIF;

between 2.052 and 3.714 across all factors), which showed that multicollinearity was not present. The residual plots (standardised residuals vs standardised predicted values) showed the data were scattered, which suggested that the assumption of homoscedasticity was met for all factors. The probability-probability (P-P) plots revealed that the residuals were normally distributed. Each factor was evaluated for extreme cases (outliers) with Cook's distance and Mahalanobis distance. An inspection of the Cook's distance revealed no outliers; however, four outliers were identified through the examination of the Mahalanobis distance (which were the same across five factors) and any values above 14.07 (with seven predictors at $p < 0.05$) were removed from further analyses (Field, 2013).

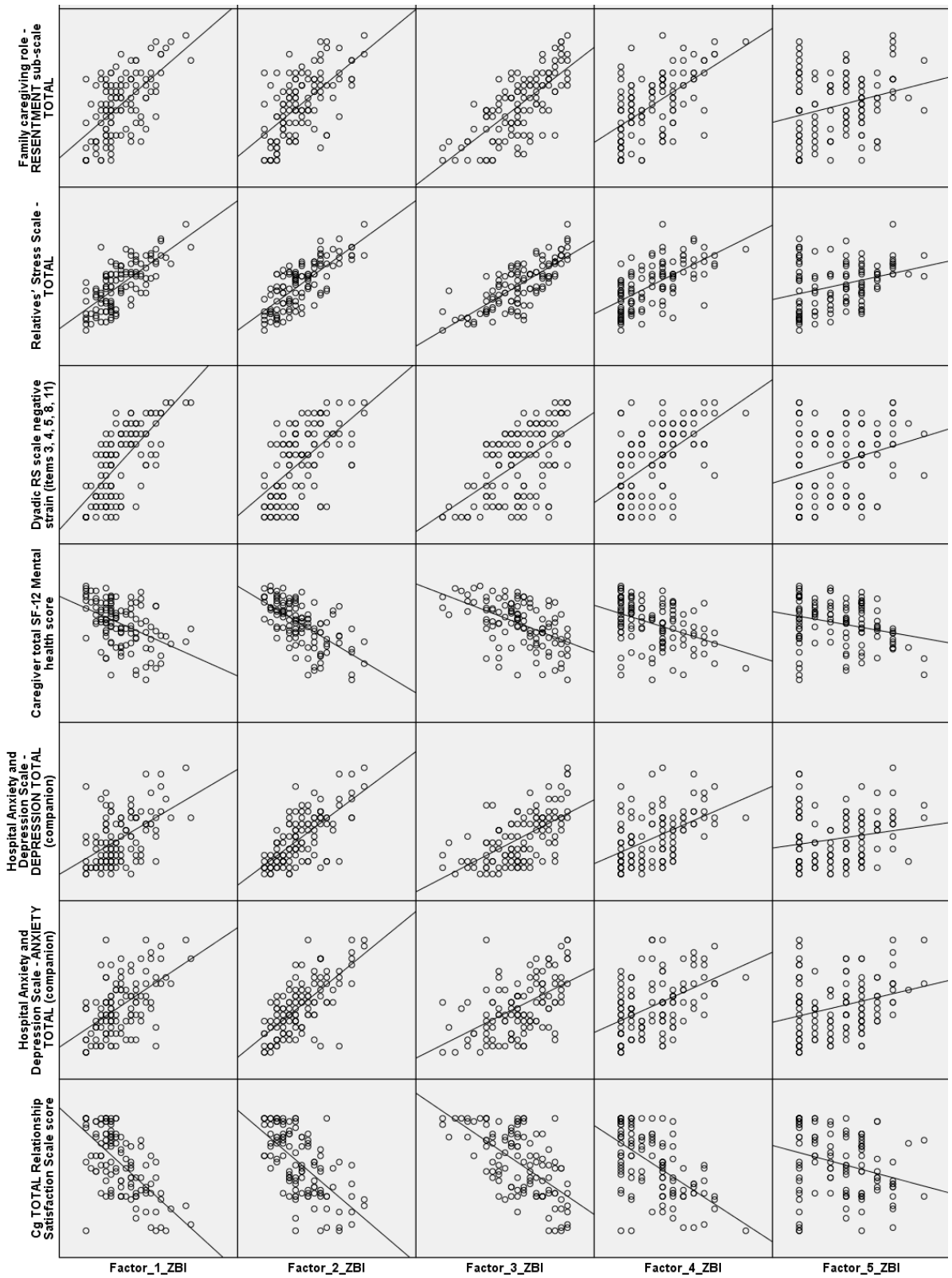
Table H.1 Assumption tests of multiple linear regression analysis for each factor.

	Factors				
Assumptions	**1**	**2**	**3**	**4**	**5**
1. Sample size (n ≥ 111)	✓	✓	✓	✓	✓
2. Linearity between IV & DV's	✓	✓	✓	✓	✓
3. Independence of residuals (Durbin-Watson test, suggested range: 1.592 – 1.807)	1.640	1.923	1.866	1.755	2.032
4. Lack of multicollinearity					
(a) correlation matrix (r < 0.8)	(a) ✓	(a) ✓	(a) ✓	(a) ✓	(a) ✓
(b) tolerance (suggested range 0.2 – 0.9)	(b) ✓	(b) ✓	(b) ✓	(b) ✓	(b) ✓
(c) VIF (< 10)	(c) ✓	(c) ✓	(c) ✓	(c) ✓	(c) ✓
5. Homoscedasticity (residual plots)	✓	✓	✓	✓	✓
6. Normal distribution of residuals (P-P plot)	✓	✓	✓	✓ *(little variance)*	✓
7. Outliers:					
(a) Cook's distance (max 1.00)	(a) 0.071 ✓	(a) 0.140 ✓	(a) 0.111 ✓	(a) 0.101 ✓	(a) 0.096 ✓
(b) Mahalanobis distance (max 14.07)	(b) 4 values > 14.07	(b)[a]	(b)[a]	(b)[a]	(b)[a]

Notes: [a] identical values identified as outliers as in factor 1

Abbreviations: IV – independent variable; DV – dependent variable; VIF – variance inflation factor

Linearity of dependent and independent variables:



Appendix I: Study 3 documents (Chapter 7)

Interview schedule

- Please could you tell me a little bit about your relationship with (name) before he was diagnosed with Parkinson's disease (PD) and/or Dementia with Lewy bodies (DLB)?
    - How would you describe it? What were the things you liked doing together? How satisfied were you with your marriage/partnership before (name's) diagnosis?
- Could you describe your satisfaction with your marriage/partnership now?
    - Has anything changed? Do you ever wish things were different in your marital relationship? Do you feel a sense of commitment and responsibility to (name)?
- How has PD/DLB affected your relationship with (name)?
    - Have you become closer or further from each other because of PD/DLB? Is your relationship stronger or weaker because of PD/DLB?
- What aspects of your life has PD/DLB affected the most?
    - What happened when (name) was diagnosed with it? What feelings were you experiencing? What changes came about? What was the hardest part of (him/her) having PD/DLB? What did you do? How did you cope?
    - What adjustments did you need to make? For example: time, energy, social support, quality of life, finances, socialising, friends, relationships, work-life and workload, hobbies, interests.

- Has intimacy changed in your relationship as a result of PD/DLB?
- *Recreational intimacy*: What social activities with (name) do you enjoy?
- *Social intimacy and support*: What type of support do you receive as a couple from other people (e.g. your friends, family, health professionals, etc)?
- *Emotional intimacy*: Does your partner listen to you when you need someone to talk to? Are you satisfied with the level of communication in your relationship? Are you able to count on (name) in times of need? Would you say your partner understands your feelings (positive and negative) most of the time? Do you ever feel lonely or distant from (name) when you are together?
- *Physical intimacy*: Would you say you are satisfied with the level of physical closeness and contact in your relationship (for example hugs, caresses, holding hands, kisses, cuddles)?
- *Intellectual intimacy*: What views do you and your partner share that are similar or dissimilar? What do you talk about?

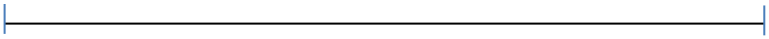
Visual analogue scale

Today's date: _____ Participant ID: ___

How would you rate your relationship satisfaction?

Place a vertical mark on the line below to indicate.

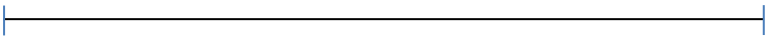
(a) Before the diagnosis of Parkinson's disease / Dementia with Lewy Bodies?



0 10

Very unsatisfying Very satisfying

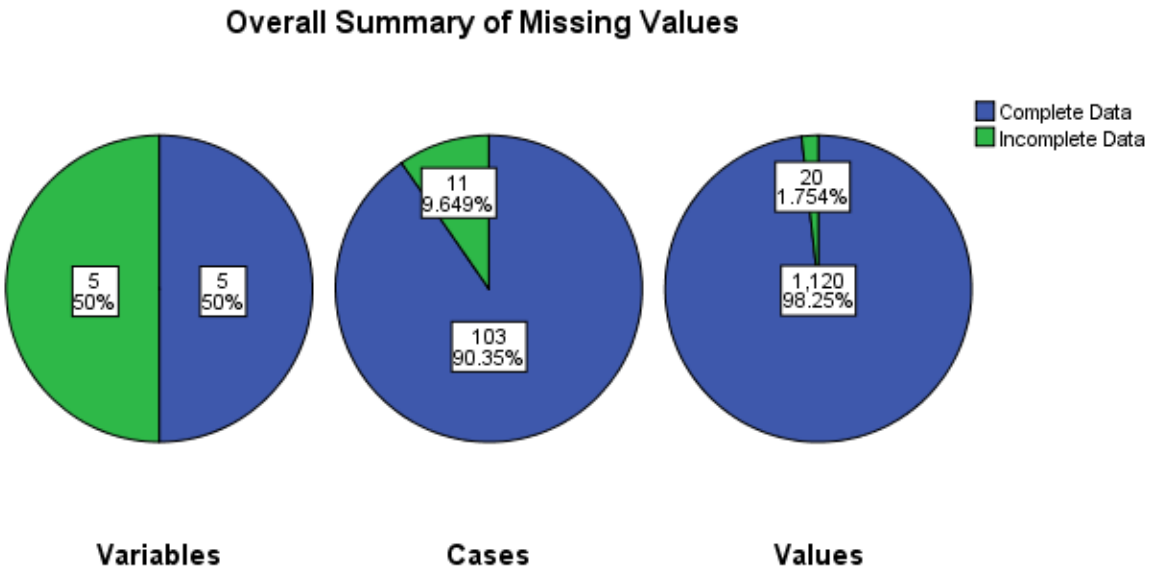
(b) Now?

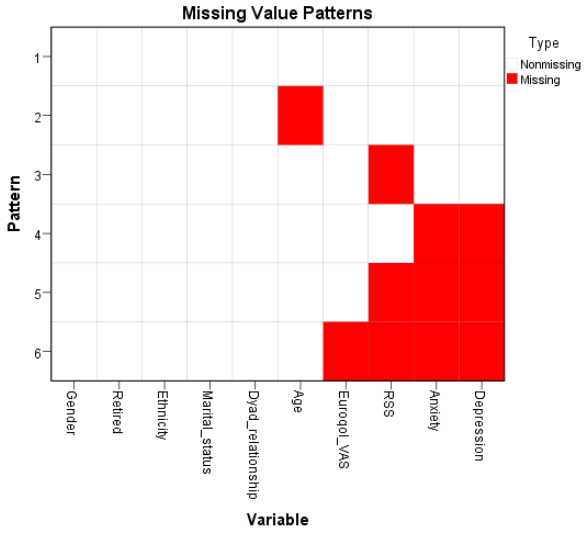


0 10

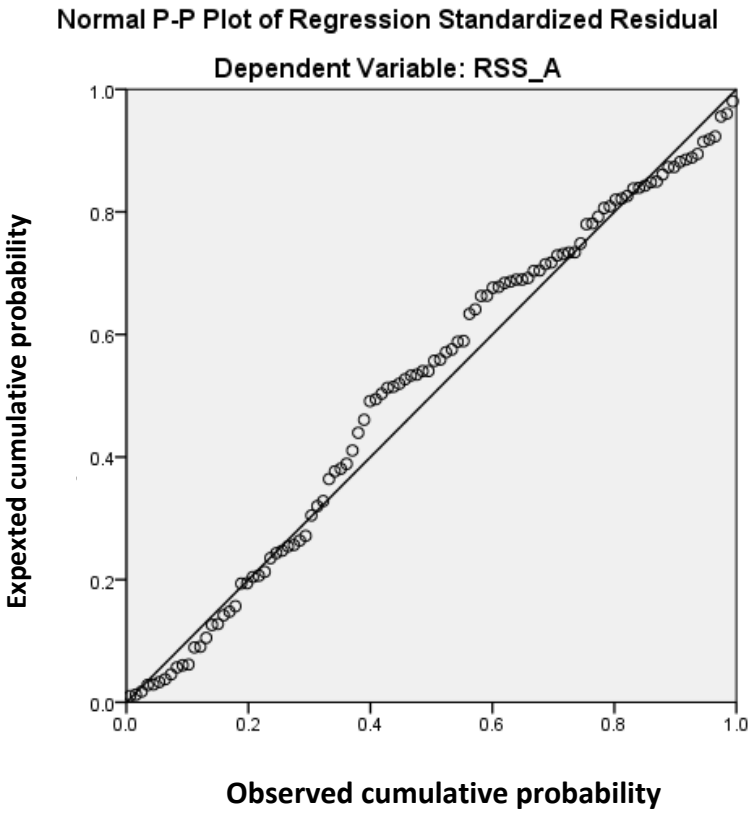
Very unsatisfying Very satisfying

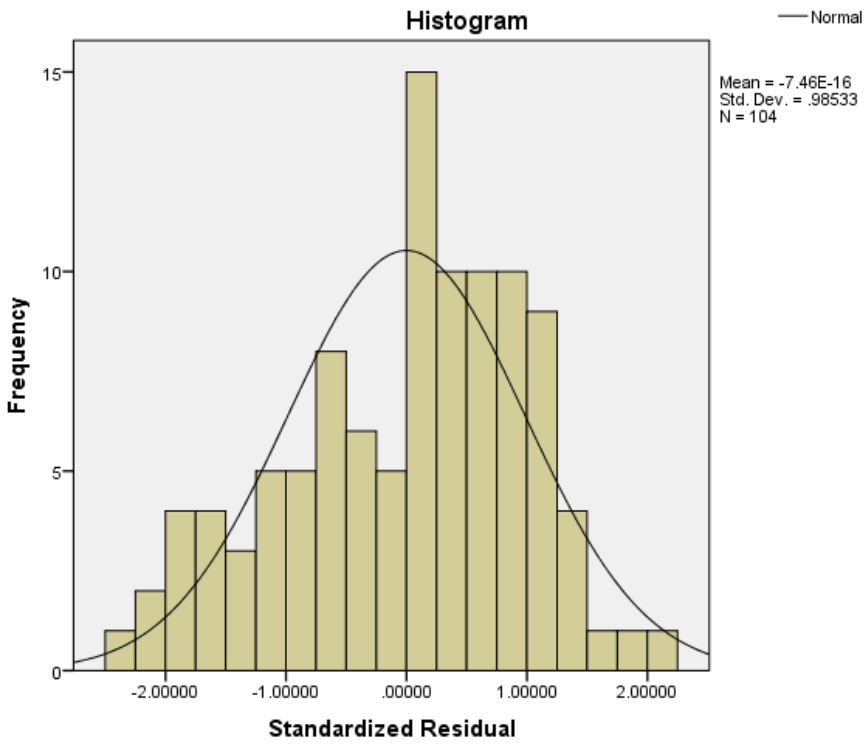
Appendix J: Study 4 documents (Chapter 8)





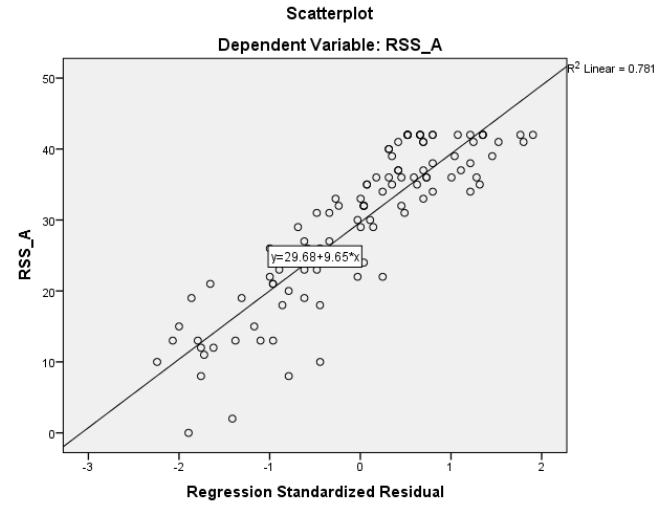
Normality


Normal P-P Plot of Regression Standardized Residual
Dependent Variable: RSS_A
Expexted cumulative probability
Observed cumulative probability
0.0
0.2
0.4
0.6
0.8
1.0

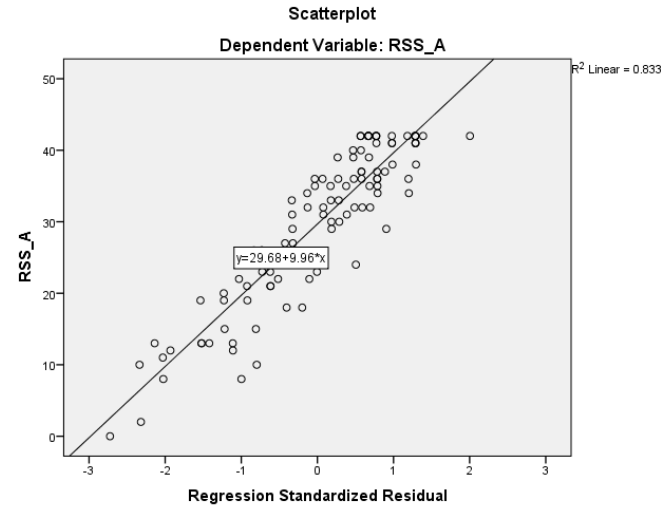

Histogram
Normal
Mean = -7.46E-16
Std. Dev. = .98533
N = 104
Frequency
Standardized Residual
0
5
10
15
-2.00000
-1.00000
.00000
1.00000
2.00000

Homoscedasticity

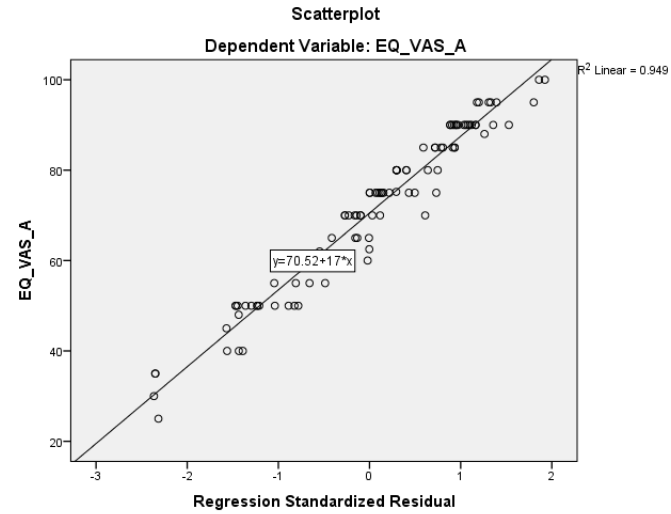
RSS (DV) & HADS depression (IV)



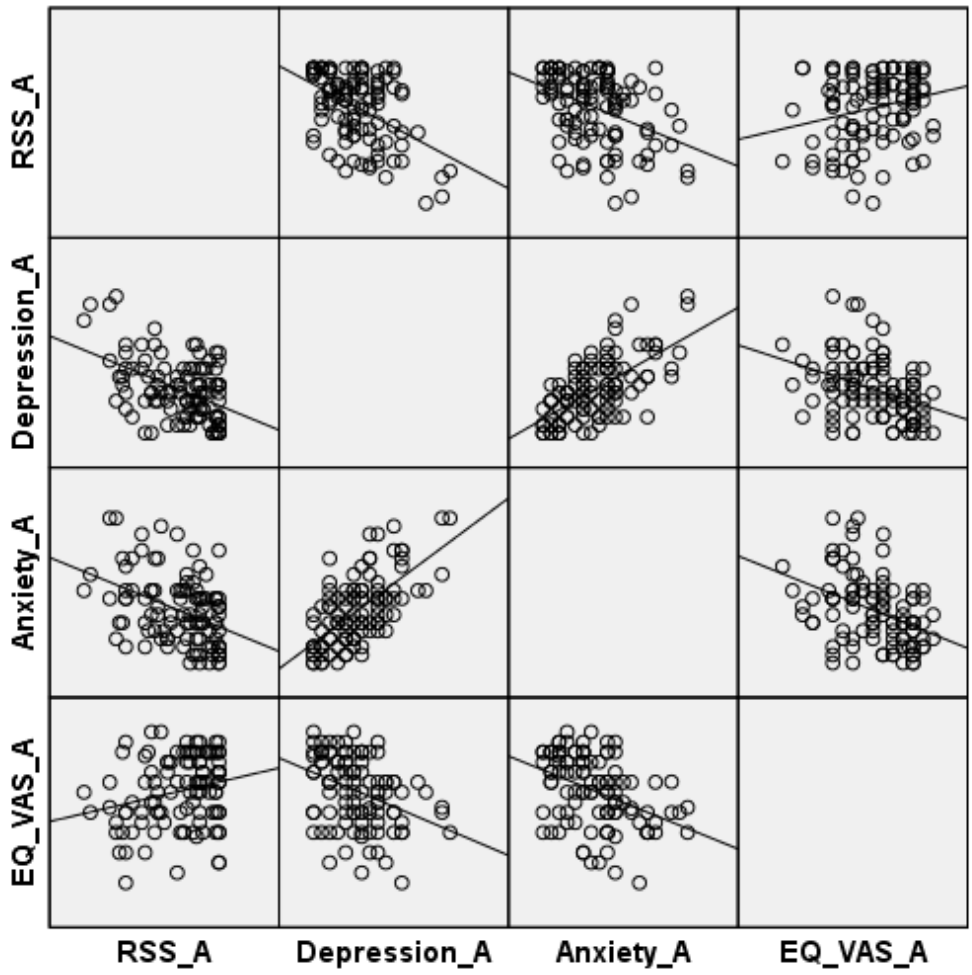
RSS (DV) & HADS anxiety (IV)



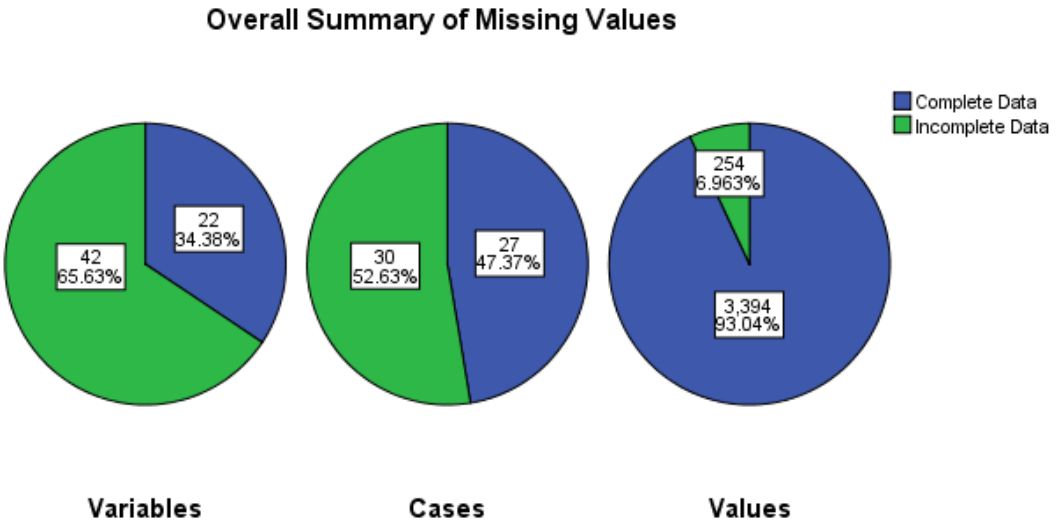
EQ-5D VAS (DV) & RSS (IV)

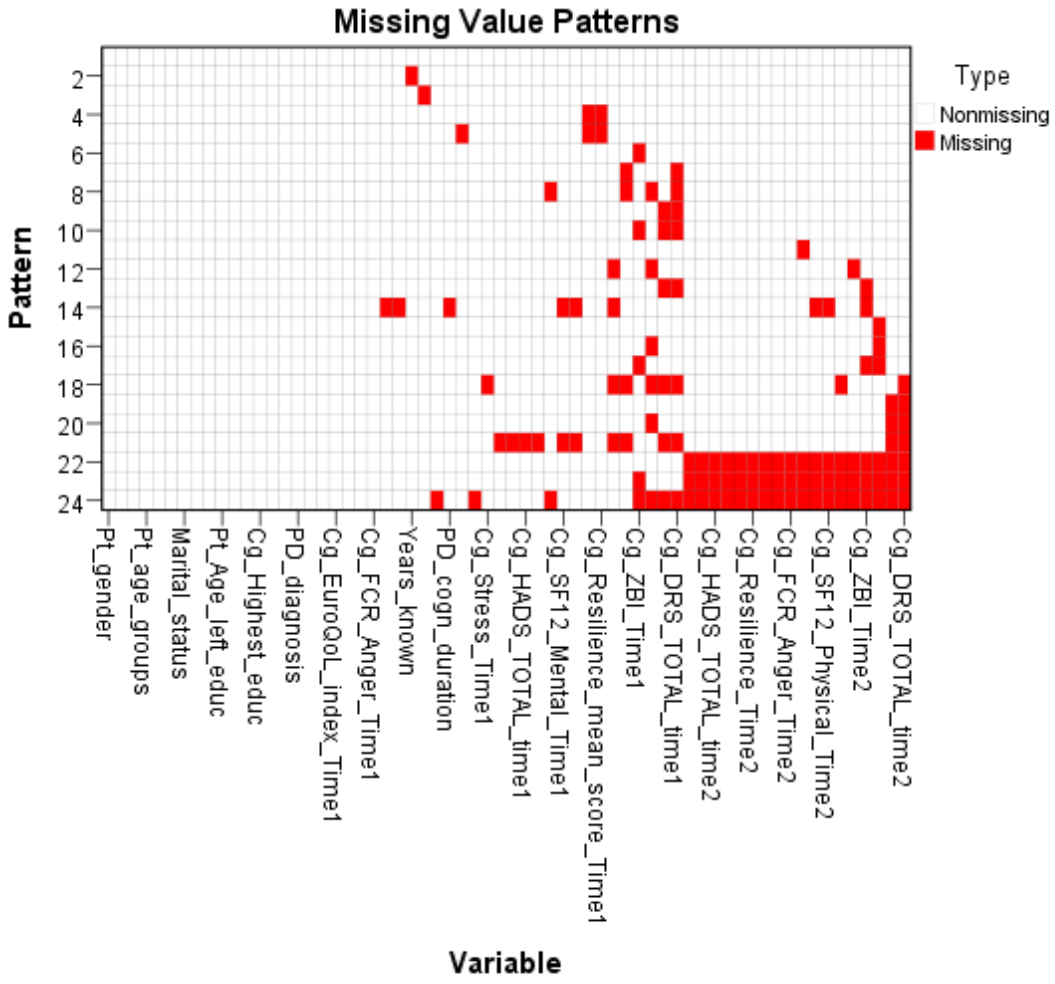


Linearity: scatterplot matrix



Appendix K: Study 5 documents (Chapter 9)





Methodological description of the assumption tests:

Prior to undertaking ANCOVA, the following assumptions were evaluated for each outcome measure via statistical tests and visual inspection of the graphs:

(1) Assumptions of **variables**:
    a. Dependent variable: Is it measured on an interval or ratio level (i.e. continuous scale)?
    b. Covariate: Is it measured on an interval or ratio level (i.e. continuous scale)?
    c. Independent variables: Do they consist of at least two independent groups (i.e. categorical data)?

(2) Assumptions of **independence**:
    a. In the sample: Are the observations (sample) random and independent (i.e. a between-subject design)? (Confirmed by randomisation whereby participants were randomly allocated either to CST-PD or TAU and participants were different in both groups)
    b. Between the independent variable (treatment effect) and the covariate (baseline outcome): Are the independent variable and the covariate independent of each other? (Checked with independent samples t-test)

(3) **Linearity** between the covariate and the dependent variable for each level of the independent variable: Is the relationship linear between the covariate and the dependent variable at each level of the independent variable? (Observed in the grouped scatterplots where the dependent variable is plotted against the covariate and grouped according to the independent variable)

(4) **Homoscedasticity of residuals:** Is the variation in the residuals constant within each randomisation group? (Assessed with residual plots)

(5) Assumptions of **homogeneity**:
    a. Variances: Is the variance in the groups equal? (Assessed with Levene's test for homogeneity of variances)
    b. Regression slopes: Are the covariate coefficients (the regression lines) parallel for each group? (Observed in scatter plot and an F-test; there should not be an interaction between the covariate and the independent variable)

(6) **Normal distribution of residuals**: Are the residuals approximately normally distributed for each group of the independent variable? (Observed with the Shapiro-Wilk test for normality, histograms, skewness and kurtosis; the allowed z-score range for skewness and kurtosis is ±1.96; Kim, 2013; some violations of normality may be tolerated unless the data is extremely skewed)

(7) **Outliers**: Are there any extreme values (outliers) in the residuals? (Viewed in box plots.)

Assumption tests for analysis of covariance (ANCOVA)

The first assumption, which set out that the dependent variable (outcome measure at follow-up) and the covariates (outcome measure at baseline) should be continuous data and the independent variables categorical data (group allocation) was confirmed for all outcome measures. All participants were randomly allocated either to CST-PD or TAU groups which confirms that the sample was independent and no participants were in two arms simultaneously (assumption 2a). An independent samples t-test was performed to determine independence between the independent variable (randomisation) and the covariate (baseline scores), which confirmed that all independent variables were unrelated to the covariates and the assumption of independence was met (assumption 2b). A linear relationship was found between baseline (covariates) and follow-up scores (dependent variable) for both randomisation arms (independent variable), as observed in the scatterplots (see below) (assumption 3).

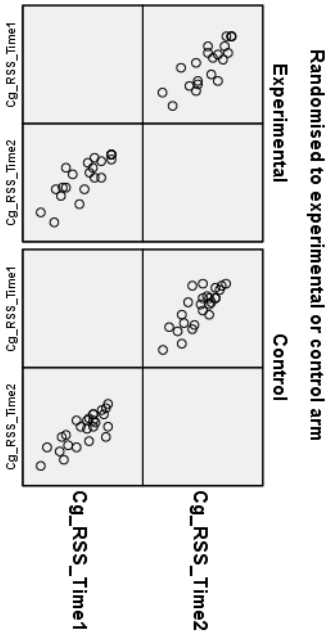
The visual inspection of scatterplots, whereby the standardized residuals were plotted against the predicted values, revealed that there was homoscedasticity on all outcome variables (assumption 4). According to Levene's test for homogeneity of variances (assumption 5a), the groups had equal variance ($p > 0.05$) for all outcome variables meeting the assumption of homogeneity of variances. Homogeneity of regression slopes (assumption 5b) was evaluated through a scatterplot and an F-test whereby the interaction between the independent variables and the covariates was assessed. A visual inspection of the scatterplots indicated that the slopes of the regression lines were parallel for all outcomes confirming that the homogeneity of regression slopes assumption was met. Additionally, the interaction term was not statistically significant for these outcomes confirming that there was homogeneity of regression slopes.

Four variables had normal distributions of standardized residuals (i.e. errors of prediction) (assumption 6) for the CST-PD arm but not for the TAU arm according to the Shapiro-Wilk test ($p < 0.05$). The standardized residuals of the SF-12 mental health sub-scale appeared to be normally distributed for the TAU group but not for the CST-PD group. Two variables (EQ-5D index and VAS scores) appeared to be kurtotic (i.e. above the allowed range of 1.96) although a visual inspection of histograms did not reveal noticeable differences between CST-PD and TAU groups. Although Shapiro-Wilk indicated one of the arms was non-normally distributed, it was decided that ANCOVA would be an appropriate statistical test for the variables as the analysis is considered relatively robust to some degree of violation of normality (Barrett, 2011; Rheinheimer & Penfield, 2001) and the variables were not

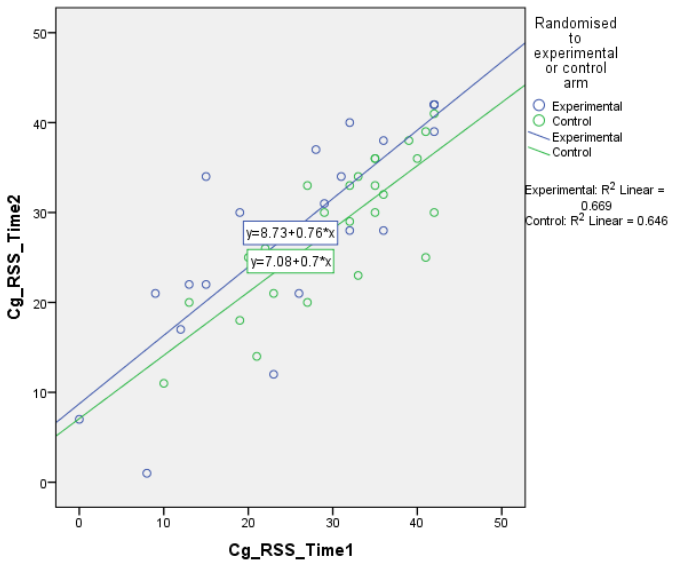
extremely skewed (i.e. within ±1.96). The data revealed that there were two outliers (assumption 7) in the 'EQ-5D index' variable as observed through visual inspection of boxplots. These outliers were reviewed in the dataset to verify that they were not due to data entry errors; rather, they were low values compared to the rest of the sample. In order to observe whether the outliers affected the output, the analyses were run with and without the outliers in the data and results were compared.

Relationship Satisfaction Scale (RSS) assumptions

Linearity of Baseline RSS & Follow-up RSS between CST-PD and TAU



Homogeneity of regression slopes (have to be fairly parallel)



Homoscedasticity

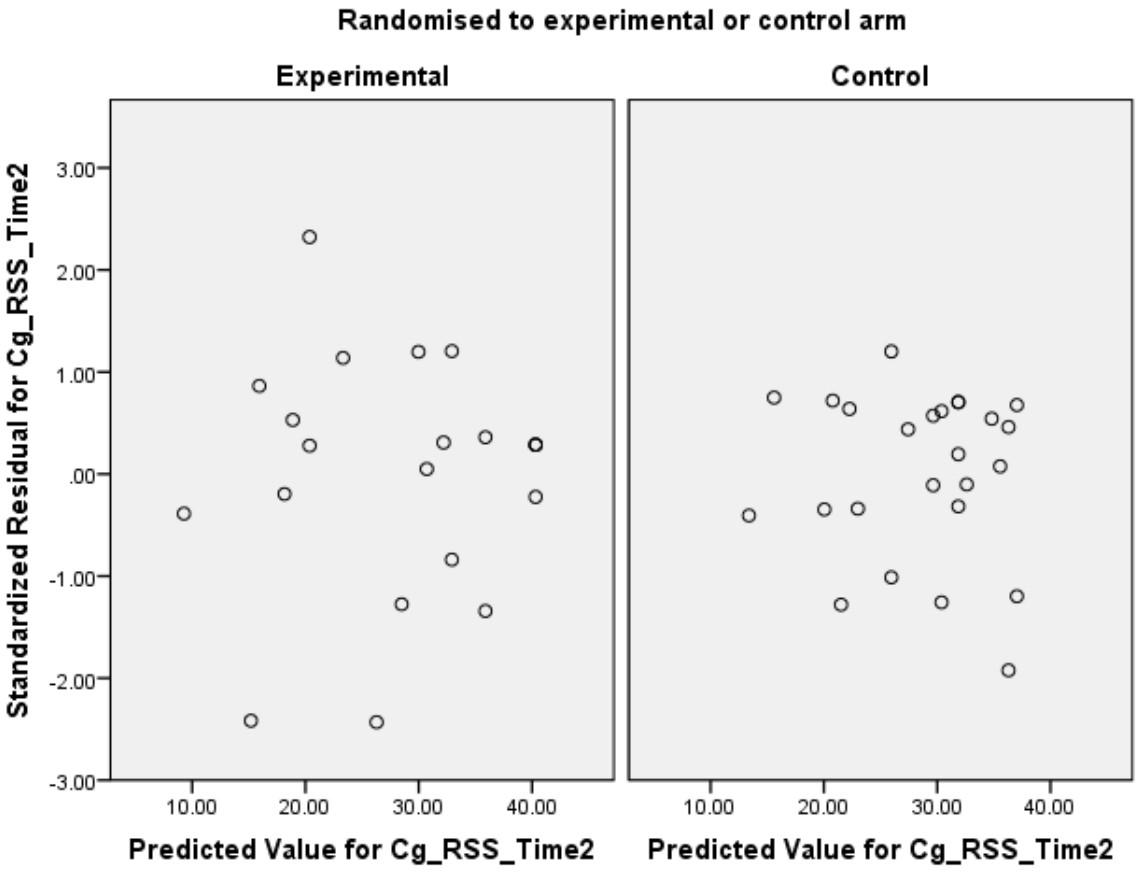


Table K.1 Assumption tests for analysis of covariance (ANCOVA).

Assumptions	RSS	DRS pos inter.	ZBI	SF-12 MCS	EQ-5D index	EQ-5D VAS
1. Types of variables						
(a) DV	(a) ✓	(a) ✓	(a) ✓	(a) ✓	(a) ✓	(a) ✓
(b) covariates	(b) ✓	(b) ✓	(b) ✓	(b) ✓	(b) ✓	(b) ✓
(c) IV's	(c) ✓	(c) ✓	(c) ✓	(c) ✓	(c) ✓	(c) ✓
2. Independence						
(a) in the sample	(a) ✓	(a) ✓	(a) ✓	(a) ✓	(a) ✓	(a) ✓
(b) between IV & covariate	(b) ✓	(b) ✓	(b) ✓	(b) ✓	(b) ✓	(b) ✓
3. Linearity between covariate & DV	✓	✓	✓	✓	✓	✓
4.Homoscedasticity of residuals (scatterplot)	✓	✓	✓	✓	✓	✓
5. Homogeneity	(a)✓	(a) ✓	(a)✓	(a)✓	(a)✓	(a)✓
(a) variances (Levene's test p-value)	p = 0.261	p = 0.872	p = 0.759	p = 0.544	p = 0.061	p = 0.797
(b) regression slopes	(b) ✓ F(1,42) = 0.116, p=0.735	(b) ✓ [F(1,39) = 0.004, p = 0.952]	(b) ✓ F(1,38) = 0.013, p=0.909	(b) ✓ F(1,41) = 1.227, p=0.274	(b) ✓ F(1,43) = 1.621, p=0.210	(b) ✓ F(1,42) = 0.073, p=0.788
6. Normal distribution of residuals *(pairwise deletion)*	(i) **x** 0.241/ 0.024	(i)✓0.860/ 0.793	(i) **x** 0.915/ 0.042	(i) **x** 0.018 / 0.349	(i) **x** 0.575/ 0.003	(i) **x** 0.346/ 0.004
(i) Shapiro-Wilk CST-PD/ TAU p-value	(ii) TAU neg. skewed	(ii) ✓	(ii) TAU pos. skewed	(ii) CST-PD neg. skewed	(ii) TAU neg. skewed	(ii) TAU neg. skewed
(ii) Histograms	(iii) TAU skewness = -0.817 ✓	(iii) ✓	(iii) TAU skewness = 1.057 ✓	(iii) CST-PD skewness = -1.430 ✓	(iii) TAU skewness = -1.614 ✓& kurtosis = 4.397	(iii) TAU skewness = -1.710 ✓& kurtosis = 4.351
(iii) skewness & kurtosis *(within ±1.96)*						
7. Outliers	✓	✓	✓	✓	2 outliers	✓

Abbreviations: CST-PD – Cognitive Stimulation Therapy in Parkinson's-related Dementia (experimental group); DRS – Dyadic Relationship Scale positive interaction sub-scale; DV – dependent variable; EQ-5D – EuroQoL index or visual analogue scale (VAS); IV – independent variable; RSS – Relationship Satisfaction Scale; SF-12 MCS – Short Form 12 Health Survey mental health (MCS) sub-scale; TAU – treatment as usual (TAU); VAS – visual analogue scale; ZBI – Zarit Burden Interview.