

**A meta-analysis of the prevalence of anxiety in informal dementia caregivers
and the efficacy of low-intensity Cognitive Behavioural Therapy-based
interventions for dementia caregivers**

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Thesis Portfolio Abstract

Objectives: To quantitatively synthesize both the prevalence of anxiety caseness in informal dementia caregivers, and the efficacy of low-intensity CBT-based interventions on the outcomes of anxiety, depression, burden, and distress (defined as stress/strain).

Methods: Systematic literature searches were conducted using electronic databases for published and unpublished literature. A random-effects meta-analysis was conducted using events and sample size data to obtain a pooled anxiety prevalence estimate. Four random-effects meta-analyses were conducted using pre and post intervention outcome measures to obtain pooled effects of low-intensity CBT-based intervention on anxiety, depression, burden and distress.

Results: Ten studies were included in the analysis of anxiety prevalence, resulting in a pooled estimate of anxiety prevalence at 32.1 percent (95% CI 20.6% to 46.2%, $p=0.01$). Significant heterogeneity was found, which could not be fully explored by subsequent sensitivity and subgroup analyses, due the limited number of studies. The number of studies included in the low-intensity CBT-based intervention meta-analyses differed across outcomes (anxiety $N = 5$; depression $N = 12$; burden $N = 3$; distress $N = 6$). The meta-analyses demonstrated significant reductions of all outcome variables. Small effects sizes were found for the reduction of anxiety ($g = 0.35$), depression ($g = 0.27$) and distress ($g = 0.33$). A medium effect was found for burden ($g = 0.53$).

Conclusions: Anxiety is a prevalent psychological difficulty experienced by informal dementia caregivers and should be afforded greater attention in the dementia caregiver literature. Low-intensity CBT-based interventions show initial promise in reducing anxiety, depression, burden and distress in dementia caregivers, however more research is required to establish the most important aspects of such interventions. Additional research recommendations and clinical implications are discussed. Limitations of both studies should be considered prior to generalising results.

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Chapter 1. Introduction to the Thesis Portfolio

Definition of Terms Used Throughout the Portfolio

This thesis portfolio focuses on the psychological difficulties experienced by people who informally care for a person with dementia. Informal caregivers are those who provide care for another person, but are not associated with a formal care system or organisation, such as employed professional caregivers or those working with voluntary organisations. People with dementia are often cared for by informal caregivers, and carers can experience psychological difficulties as a result of their caregiving role (Brodaty & Donkin, 2009; Friedman, Shih, Langa, & Hurd, 2015).

Throughout this thesis portfolio, the term 'dementia caregiver' refers to any person who provides informal support for a person who has a dementia illness. The term 'dementia' is an umbrella term used to describe a number of illnesses which involve the progressive decline of brain functioning. As such, dementia is differentiated from other stable or potentially reversible cognitive impairments, such as a traumatic brain injury or depression. This portfolio uses the term 'dementia caregiver' to refer to caregivers of people who have any type of dementia illness.

This portfolio does not place limits on how long and how often a person has been providing care, what caregiving duties are carried out, or any other care related variables. This is largely because there is no universal definition of what constitutes being a caregiver. In this portfolio, therefore, it was decided to use a broad definition of what constitutes a 'caregiver' (i.e., not excluding caregivers based on the above mentioned variables) to avoid the risk of excluding relevant evidence. Instead, the potential factors that may affect the outcomes of interest will be discussed throughout.

Selecting a narrower definition of 'caregiver' (e.g. Western female caregivers aged 65 years and over) may increase specificity, however it would significantly reduce the generalisability of any findings. Given that dementia is a global illness and caregivers are

arguably a heterogeneous group, it is therefore important that the chosen definition of informal caregiver does not arbitrarily exclude parts of the population. Nonetheless, it is important to acknowledge the variety within in the dementia caregiver population, based on the above definition, and to explore the impact of such differences by selecting appropriate research methodology (i.e. moderator analysis).

Overview of the Thesis Portfolio

The portfolio includes two empirical papers, both of which employ a quantitative meta-analytical method, and have been written for publication in a scientific journal. Following the first empirical paper, there is a bridging chapter which provides a clear rationale for linking the two papers. An overall discussion chapter is included, which provides suggestions for future research and clinical implications to improve support for caregivers of people with dementia. The references are listed, and appendices are provided.

Chapter 2. Empirical Paper One

This chapter consists of a systematic review and meta-analysis, written for publication in the journal *International Psychogeriatrics*. This chapter is formatted in accordance with the journal submission guidelines (Appendix A).

Abstract word count: 244 (250 words journal limit)

Total word count (excluding abstract, tables, figures and appendices): 4998 (5000 words journal limit)

Running Head: ANXIETY PREVALENCE IN DEMENTIA CAREGIVERS

Anxiety in informal dementia caregivers: a meta-analysis of prevalence

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Abstract

Objectives: This study aimed to quantitatively review the current prevalence of anxiety caseness in informal caregivers for people with a progressive dementia illness.

Design: Literature searches were conducted in databases of published (PsycINFO; MEDLINE; CINAHL; Scopus) and unpublished literature (Open Grey; ProQuest). Events and sample size data was pooled using a random-effects model meta-analysis to obtain a summary prevalence percentage.

Included studies: Studies reporting on current anxiety caseness in adult informal caregivers for people with any progressive dementia illness were included. Studies were excluded if they reported lifetime or duration of caregiving anxiety prevalence only, or if only mean anxiety scores were reported.

Measurements: Anxiety caseness was defined as the presence of any anxiety disorder using a reliable and valid anxiety diagnostic tool or the presence of a clinical level of anxiety symptoms, as assessed using a reliable and valid self-report symptom measure with a clinical cut-off score.

Results: A total of 10 studies were included, resulting in a pooled estimate of anxiety prevalence at 32.1 percent (95% CI 20.6% to 46.2%, $p=0.01$). Significant heterogeneity was found, which was not reduced following sensitivity analysis. Subsequent subgroup analyses were limited by the number of studies included, and the characteristics of included studies.

Conclusions: This study suggests anxiety is a prevalent difficulty experienced by informal adult dementia caregivers. However, the ability to draw meaningful conclusions regarding potentially relevant moderating factors was limited by the small number of included studies. Recommendations for future research are discussed.

Keywords

Dementia caregivers; anxiety; psychological difficulties; prevalence

Introduction

Dementia is an umbrella term used to describe a number of illnesses resulting in progressive cognitive decline. Different dementia illnesses can have differing profiles of difficulty, with changes occurring in areas such as memory, attention, mood and personality (Johnson *et al.*, 2011). People who have dementia are often cared for by informal caregivers, who are usually relatives or friends of the cared for person (Friedman *et al.*, 2015).

Providing informal care for a person with dementia can be challenging for caregivers. Caregiving often involves providing practical support with daily living tasks, in addition to emotional support and assistance in areas such as communication and decision making (Alzheimer's Research UK, no date). Caregivers are also often required to provide support in the context of changes in personality and behaviours which challenge. Such behaviours can include wandering, shouting, physical aggression towards the caregiver, and destruction of personal possessions (Andrews, 2006). Furthermore, caregiving is time consuming and carers often become socially isolated (Brodaty and Donkin, 2009). Caregiving can also place demands on a caregiver's financial resources, as carers may incur additional costs related to caregiving or may have to reduce time spent in employment to attend to the cared for person's needs (Janssen, 2013).

Whilst there are reports of positive aspects to caregiving, the literature typically demonstrates a negative impact on the psychological wellbeing of caregivers (Roff *et al.*, 2004). Difficulties include clinical levels of depression and anxiety, increased levels of burden and stress, as well as reduced life satisfaction (Dahlrup *et al.*, 2015; Schulz *et al.*, 1995; Sörensen & Conwell, 2011). Such difficulties are notable compared to both the non-caregiver population, and to caregivers of people with non-dementia illnesses (Brodaty and Donkin, 2009; Mausbach *et al.*, 2013). Although a negative impact on the psychological wellbeing of caregivers of people with dementia can be broad, much of the

literature has focused on depression and burden as primarily outcomes, and anxiety appears somewhat neglected. Yet it has been suggested that the majority of depressed caregivers also experience comorbid anxiety, as well as anxiety occurring independent of depression in other caregivers (Mahoney *et al.*, 2005).

This begs the question why has comparatively less attention been afforded to anxiety in dementia caregivers compared to depression. One hypothesis is that the prevalence of anxiety is less reported in the literature. Indeed, a previous review of the prevalence of anxiety in dementia caregivers found only four studies which measured anxiety diagnoses and caseness level symptoms using valid measurement tools (Cooper *et al.*, 2007). A more recent review and meta-analysis again found only four studies reporting prevalence though the study focused only on caregivers for people with Alzheimer's dementia (Sallim *et al.*, 2015). By comparison, a recent meta-analysis found 38 studies examining the prevalence of depression in dementia caregivers (Collins and Kishita, 2018).

It is therefore important that the prevalence of anxiety in dementia caregivers is established, as doing so may help shape future caregiver literature. In addition, several factors are postulated to have an impact on caregiver wellbeing, including caregiver variables (e.g. gender, ethnicity, coping style), cared for person factors (e.g. severity of impairment) and environmental factors (e.g. culture; country development status) (Janevic and Connell, 2001; Covinsky *et al.*, 2003; Shaji, 2009; Wong *et al.*, 2012; Snyder *et al.*, 2015). Therefore, it is important to determine if anxiety is a prevalent difficulty worth examining further in terms of understanding potential mediating and moderating influences. Furthermore, if anxiety is a prevalent difficulty in the dementia caregiver population, the inclusion of anxiety as an outcome measure in intervention studies will be further justified. This will ensure intervention studies focus on all relevant outcomes and provide a more valid estimate of intervention effectiveness.

Therefore, this study aims to provide a synthesized estimate of the prevalence of anxiety, defined as anxiety diagnoses or caseness level of symptoms, in the dementia caregiver population.

Method

Protocol and Registration

The review protocol was published on the PROSPERO international prospective register of systematic reviews (registration number: CRD42018087895; accessed via www.crd.york.ac.uk/PROSPERO).

Search Strategy

A systematic search of published literature was conducted using the electronic databases PsycINFO, MEDLINE, CINAHL and Scopus. A search of unpublished literature was conducted to address potential publication bias, using Open Grey and ProQuest. Reference lists of key review papers were hand searched. Key search terms included (a) terms related to the cared for person illness (dementia OR Alzheimer* OR "Lewy body" OR "fronto*temporal"). Cared for person illness terms were selected with the aim of capturing caregivers of all types of dementia. It was expected that the umbrella term "dementia" would capture most of the relevant studies, including well-known and lesser known types of dementia. However, additional terms relating to the most prevalent dementia illnesses were also included. "Vascular" was not included as a term as it was expected that this term would identify studies relating to non-dementia vascular illnesses. (b) Terms relating to caregivers (caregiver* OR carer*) were included to identify studies which focused on caregivers as opposed to the person with dementia. (c) A single term "anxiety" was used to identify studies which focused on clinical levels/diagnoses of anxiety. Other terms were considered, such as "worry", however it was expected that such terms would decrease specificity to an unacceptable level. (d) Terms relating to the

language used to describe studies which report on prevalence were included (prevalence OR preval* OR epidemiol* OR “presence of”). Limits were placed for English language publications. Sources were searched from the date of database inception to December 31st 2017. **Eligibility Criteria**

Articles were eligible if the following criteria were met: (a) Participants are adult informal caregivers (may receive financial support as part of caregiving role, e.g. government benefits, but must not be formal paid caregivers, e.g. ‘professional care assistant’); for a person with a progressive dementia illness (b) the number of participants and current anxiety caseness prevalence rate is reported; (c) anxiety caseness is assessed as the presence of any anxiety disorder using a reliable and valid anxiety diagnostic tool OR the presence of a clinical level of anxiety symptoms, as assessed using a reliable and valid self-report symptom measure with a clinical cut-off score; (d) the study is reported in English. Exclusion criteria included: (a) participants include non-dementia caregivers; (b) reports incidence only; (c) reports sample mean symptom scores only; (d) reports lifetime or duration of caregiving prevalence only.

Restrictions were not placed on caregiver demographic details or characteristics as this study aimed to assess the prevalence of anxiety across the whole population of dementia caregivers.

Selection of Studies

The primary reviewer (LK) conducted the initial search and duplicates were excluded. Potentially relevant articles were identified based on title and abstract. Full articles were obtained and assessed for eligibility by LK. The secondary reviewer (NK) assessed eligibility for ten percent of articles to reduce bias in the selection procedure. Discrepancies were discussed and resolved, and a third reviewer was available for consultation however this was not required.

Quality Assessment and Risk of Bias

The quality and risk of bias of included studies was assessed using a the Prevalence Critical Appraisal Instrument (PCAI) which is designed specifically for assessing prevalence studies (Munn *et al.*, 2014). Whilst other relevant quality assessment tools exist, the PCAI focuses on assessing the quality of the study methods that were planned and carried out, rather than assessing the quality of the written report, which can be misleading (Harder, 2014). The PCAI consists of ten items regarding the internal and external validity of a study. Each item is rated as either 'yes', 'no', or 'unclear'. Furthermore the tool includes a comprehensive usage guide (Munn *et al.*, 2014). The items regarding the use of a sample representative of the target population, the description of participants and setting, and the identification of confounding factors and subgroup differences were further defined to ensure consistent rating for this study (Appendix B).

The PCAI does not provide a quantitative quality level for studies. It was decided that all items on the tool were of equal importance for this review, therefore a study was defined as poor quality if the total number of 'yes' items achieved was less than half.

All articles were assessed for quality by LK and ten percent of articles were assessed by NK to reduce bias. Discrepancies were discussed and resolved. Articles defined as low quality were selected for exclusion in sensitivity analysis.

Data Extraction

An electronic form was used to extract study characteristics, participant information, prevalence measurement tool characteristics and prevalence data, as per Cochrane guidelines (Higgins and Green, 2011) (Appendix C). The data was extracted by LK for all articles, NK extracted data for ten percent of the articles. Data extraction was considered reliable when there were no discrepancies found between the two reviewers.

Statistical Analysis

Statistical analyses were performed using Comprehensive Meta-Analysis (CMA) software (Biostat Inc., 2014). The analysis used one-group events and sample size data to calculate a pooled prevalence estimate. A random-effects model was selected to pool the data, due to expected variation in participant characteristics and prevalence measurement tool. The random effects model is arguably the most appropriate model as it assumes each study contains its own variance as a result of variation in study characteristics (Borenstein *et al.*, 2010).

Heterogeneity and Subgroup Analysis

Heterogeneity was examined visually using a forest plot and the calculation of the I^2 statistic, which shows the percentage of the total variance which can be explained by heterogeneity (Cuijpers, 2016). Planned sensitivity analysis included the removal of outliers, studies rated as poor quality, and studies which appeared to meet the inclusion criteria but contained uncertainty. Remaining heterogeneity was planned to be explored through subgroup analysis using the following moderators: (a) prevalence measurement tool type (diagnostic or self-report); (b) specific prevalence measurement tool; (c) care-recipient dementia type (only where study sample contains a heterogeneous group of care-recipients based on dementia type); (d) country development status, defined as the Human Development Index (HDI) category (low; medium; high; very high) which was determined based on the study country for the purposes of this review (United Nations Development Programme, 2016). Potentially relevant moderators using summary data (e.g. duration/hours caregiving) were not examined due to potential aggregation bias (Kaufmann *et al.*, 2016).

Publication Bias

Publication bias was explored visually using a funnel plot, and the 'trim and fill' method was applied to estimate prevalence after bias had been taken into account (Borenstein *et al.*, 2009; Duval and Tweedie, 2000). Rosenthal's Fail-safe N was

calculated to estimate the number of missing studies which would be required to reduce the p -value to below .05 (Rosenthal, 1979).

Results

Study Selection

The selection of studies is outlined in Figure 1. The search yielded 768 articles, of which 332 were excluded as duplicates, resulting in 436 articles which were screened based on title and abstract. Thirty-seven articles were subject to full eligibility screening, resulting in a total of ten eligible studies (Dura *et al.*, 1991; Coope *et al.*, 1995; Sansoni *et al.*, 2004; Mahoney *et al.*, 2005; García-Alberca *et al.*, 2011; García-Alberca *et al.*, 2012; Medrano *et al.*, 2014; Ostojić *et al.*, 2014; Weaving *et al.*, 2014; Ervin *et al.*, 2015). The most common reason for exclusion was due to prevalence data not being reported, with mean anxiety scores or correlations of anxiety scores with other variables being reported instead. Of the ten eligible studies, one study was identified for exclusion in sensitivity analysis (Sansoni *et al.*, 2004). Sansoni *et al.* (2004) used a higher symptom cut-off score than usual for the anxiety measurement tool used, meaning the study may have underestimated the prevalence rate compared to studies using a lower cut-off score. Furthermore, the study employed a non-typical study design as is described below.

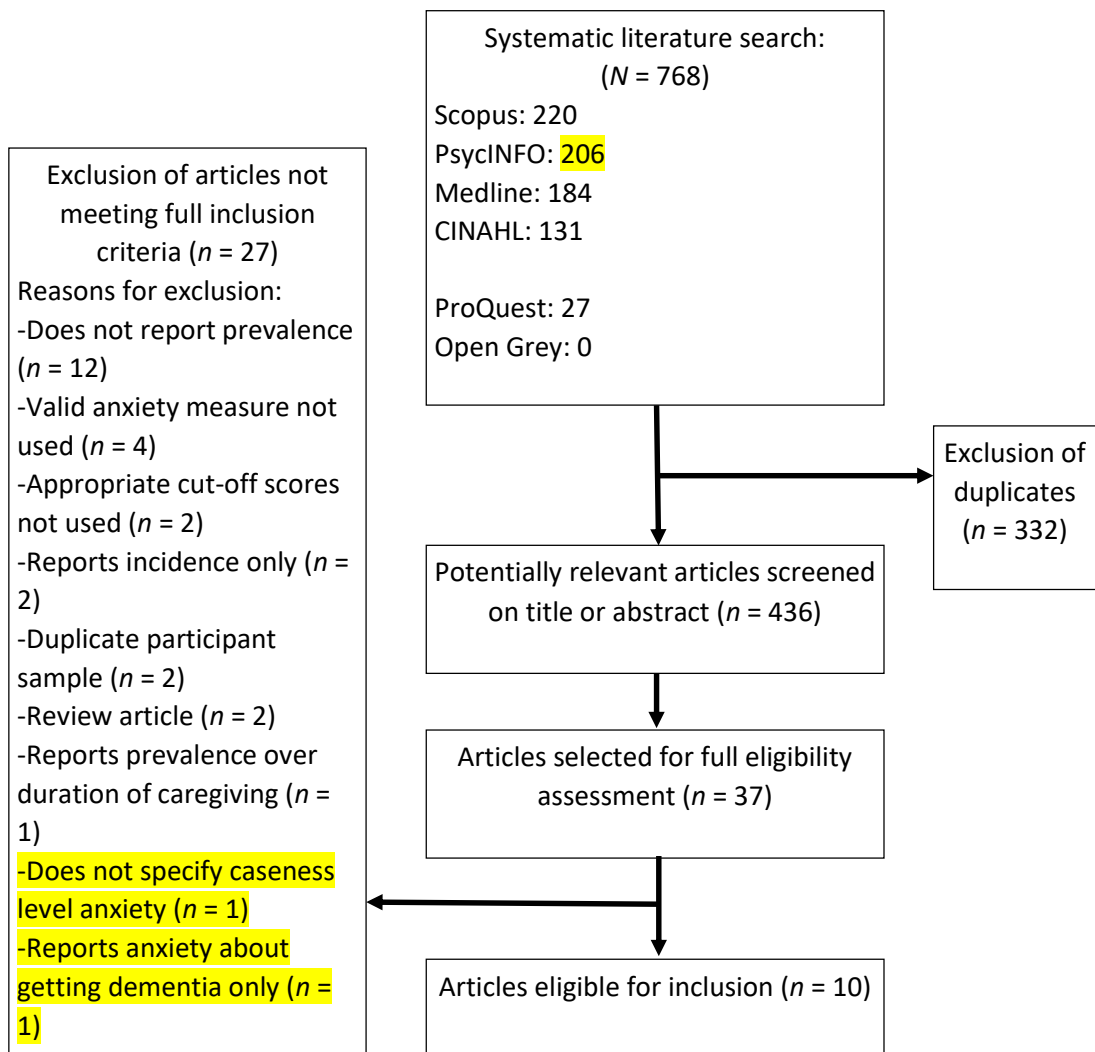


Figure 1. Systematic literature search flow chart

Study Characteristics

Participant characteristics

Participant characteristics are summarised in Table 1. The ten studies comprised a total of 918 participants, with mean ages ranging from 48.7 (SD = 10.1) to 66.7 (SD = 12.6) years. The majority of caregiver participants were female spouses or adult children of the cared for person.

Table 1. Characteristics of participants for each study

Study	Country	Total N	Mean age (SD)	% female	Relationship to care-recipient (%)	Mean hours caregiving per day (SD)	Mean number of months caregiving (SD)	Care-recipient dementia type (%)
Coope et al. (1995)	UK	109	NR	62	Spouse (42); Adult child (39.5); Sibling (4.6); Distant relative/friend (14.7)	NR	NR	Any mild or moderate Dementia (NR)
Dura et al. (1991)	USA	78	48.74 (10.11)	84.6	Adult child (100)	4.61 (4.94)	48.83 (40.22)	Alzheimer's disease (80.77); Multi-infarct dementia (3.85); Huntingdon's dementia (3.85); Parkinson's dementia (7.69); Unspecified dementia (3.85)
Ervin et al. (2015)	Australia	39	NR	77	Daughter (28); Wife (28); Husband (18); Other family member/friend (21)	NR	NR	Not specified dementia (43); Alzheimer's disease (46); Other (Parkinson's dementia, Vascular dementia, Cerebral amyloidosis) (10)
García-Alberca et al. (2011)	Spain	125	61.41 (11.03)	79.2	Adult child (44); Spouse (41.9); Sibling (6.4); Other relative (8)	NR	NR	Alzheimer's disease (100)
García-Alberca et al. (2012)	Spain	80	62.15 (10.37)	77.5	Adult child (43.8); Spouse (38.8); Sibling (7.4); Other relative (10)	NR	NR	Alzheimer's disease (100)
Mahoney et al. (2005)	UK	153	64 (3.3)	69.9	Spouse (44.4); Adult child (44.4); Friend (4.6)	NR	NR	Alzheimer's disease (NR); Dementia (NR)

Medrano et al. (2014)	Dominican Republic	67	61 (NR)	84	Adult child (55); Spouse (15); Grandchild (12); Sibling (9); Other relative (9)	NR	NR	Alzheimer's disease (100)
Ostojić et al. (2014)	Croatia	30	57.6	73.3	Adult child (63.3); Spouse (26.7)	16.43 (9.93)	NR	Alzheimer's disease (100)
Sansoni et al. (2004)	Italy	34	59.21 (9.62)	100	Wife (73.53); Sister (1.94); Daughter (11.76); Other relative (8.82); Friend (2.94)	19.38 (4.75)	47.76 (34.08)	Alzheimer's disease (100)
Weaving et al. (2014)	UK	203	66.71 (12.64)	69.8	Spouse (61.5); Adult child (33.5); Other relative/friend/partner (5)	NR	NR	NR

Note. N = Number of participants; NR = Not reported.

Measurement and design characteristics

Measurement and design characteristics are summarised in Table 2. The majority of studies used self-report symptom measures to determine anxiety caseness prevalence, with only two studies reporting anxiety prevalence based on a diagnostic tool. Study design was similar across all included studies. Eight studies employed a cross-sectional design and one study used a retrospective case-control design which also reported current cross-sectional prevalence. Another study used a descriptive repeated measures design, where the anxiety symptom measure was administered over nine weeks (Sansoni *et al.*, 2004). The authors described the design was chosen to eliminate coincidental bias, and as the study reported consistent anxiety scores over the nine-week period it was included in this review.

Table 2. Characteristics of measurements and design for each study

Study	Study design	Anxiety measurement type and tool	Cut-off point/criteria for caseness	Prevalence Caseness % (N)
Coope et al. (1995)	Cross-sectional	Diagnostic - GMS-AGECAT	Level ≥ 3	3.67 (4)
Dura et al. (1991)	Retrospective case-control incl. cross sectional data	Diagnostic - SCID-NP	Meets DSM-III-R diagnostic criteria for Generalised Anxiety Disorder, Social Phobia or Panic Disorder	7.69 (6)
Ervin et al. (2015)	Cross-sectional	Self-report symptom measure - DASS-21	≥ 10 (incl. moderate (10-14); severe(15-19); extremely severe (≥ 20))	26 (10)
García-Alberca et al. (2011)	Cross-sectional	Self-report symptom measure - STAI-S	≥ 28	53 (66)
García-Alberca et al. (2012)	Cross-sectional	Self-report symptom measure - STAI-S	≥ 28	56.6 (45)
Mahoney et al. (2005)	Cross-sectional	Self-report symptom measure - HADS-A	≥ 11	23.5 (36)
Medrano et al. (2014)	Cross-sectional	Self-report symptom measure - HARS Spanish version	≥ 6 (incl. mild 6-14; moderate/severe ≥ 15)	19 (13)
Ostojić et al. (2014)	Cross-sectional	Self-report symptom measure – HADS-A Croatian version	≥ 11	46.7 (14)

Sansoni et al. (2004)	Descriptive correlational repeated measures	Self-report symptom measure - STAI-S	>40	76.47 (26)
Weaving et al. (2014)	Cross-sectional	Self-report symptom measure - HADS	≥11	49.2 (100)

Note. NR = Not reported; GMS-AGECAT = Geriatric Mental State Schedule interview - Automated Geriatric Examination for Computer Assisted Taxonomy (Copeland *et al.*, 1988); SCID-NP = Structured Clinical Interview - non-patient version (Riskind *et al.*, 1987); DASS-21 = Depression Anxiety Stress Scales (Ng *et al.*, 2007); STAI-S = State-Trait Anxiety Inventory - state subscale (Spielberger *et al.*, 1970); HADS-A = Hospital Anxiety and Depression Scale – anxiety scale (Zigmond and Snaith, 1983); HARS = Hamilton Anxiety Rating Scale – Spanish version (Lobo *et al.*, 2002).

Study Quality and Risk of Bias

The assessment of study quality and bias using the PCAI is presented in Table 3. The quality of included studies varied, with studies achieving between three and seven positive items out of a total of ten. Three studies achieved less than five positive items and were selected for exclusion in sensitivity analysis (Sansoni *et al.*, 2004; Medrano *et al.*, 2014; Ostojčić *et al.*, 2014).

Participants and recruitment

There were no large scale national studies included. Only one study sought a participant sample which could be described as representative of the target population (Mahoney, *et al.*, 2005). All others did not seek representative samples. For example, Coope *et al.* (1995) only included caregivers of people with mild or moderate dementia, and Ervin *et al.* (2015) only included rural caregivers and excluded those which were predicted to be burdened by participating. Several studies limited recruitment based on participant characteristics such as gender (Sansoni *et al.*, 2004), cared for person dementia type (Sansoni *et al.*, 2004; García-Alberca *et al.*, 2011; García-Alberca *et al.*, 2012; Medrano *et al.*, 2014; Ostojčić *et al.*, 2014), or relationship to the cared for person (Dura *et al.*, 1991). Weaving *et al.* (2014) only recruited caregivers who accessed support from voluntary services, and did not seek to recruit caregivers who were not currently engaged in caregiver support. However, the majority of studies described appropriate recruitment methods for their chosen samples (e.g. screening using eligibility criteria), as per PCAI guidelines (Munn *et al.*, 2014).

Sample size calculations were not appropriately conducted by any of the studies. Therefore, it was necessary to calculate the minimum sample size required to detect described prevalence rates at a confidence level of 95% (Naing *et al.*, 2006). It was noted that only two studies had used an adequate sample size (Dura *et al.*, 1991; Coope *et al.*, 1995). Furthermore, several studies did not provide an adequate description of participant characteristics (Coope *et al.*, 1995; Sansoni *et al.*, 2004; Mahoney, *et al.*, 2005; García-

Alberca *et al.*, 2012; Medrano *et al.*, 2014; Ervin *et al.*, 2015). These studies neglected to describe at least six of the 12 identified important participant characteristics (Appendix B). There did not appear to be any pattern regarding which characteristics were neglected across these studies.

Data measurement and analysis

Only one study was determined to have conducted data analysis with sufficient coverage of the sample (Coope *et al.*, 1995), two studies had a high proportion of drop-outs (Sansoni *et al.*, 2004; García-Alberca *et al.*, 2012), and all other studies did not report the number of participants that declined to participate or dropped-out. All included studies used objective and reliable prevalence measurement tools, as doing so formed part of the inclusion criteria for this review. All studies were found to use appropriate statistical analysis methods. Only one study gave sufficient consideration to confounding factors and subgroups/moderators (Mahoney, *et al.*, 2005). Six studies included analysis of some moderating factors (Sansoni *et al.*, 2004; García-Alberca *et al.*, 2011; Medrano *et al.*, 2014; Weaving *et al.*, 2014; Ervin *et al.*, 2015), though the criteria used to define the moderators was not clear in three of those studies (Sansoni *et al.*, 2004; García-Alberca *et al.*, 2011; García-Alberca *et al.*, 2012; Medrano *et al.*, 2014). Three studies did not give consideration to any moderating factors (Dura *et al.*, 1991; Coope *et al.*, 1995; Ostojic *et al.*, 2014).

Table 3. Assessment of quality and risk of bias using the Prevalence Critical Appraisal Instrument for each study

Study	Item 1	Item 2	Item 3	Item 4	Item 5	Item 6	Item 7	Item 8	Item 9	Item 10	Total number of 'Yes' items
Coope et al. (1995)	No	Yes	Yes	No	Yes	Yes	Yes	Yes	No	N/A	6
Dura et al. (1991)	No	Yes	Yes	Yes	Unclear	Yes	Yes	Yes	No	N/A	6
Ervin et al. (2015)	No	Yes	No	No	Unclear	Yes	Yes	Yes	No*	Yes	5
García-Alberca et al. (2011)	No	Yes	No	Yes	Unclear	Yes	Yes	Yes	No*	Unclear	5
García-Alberca et al. (2012)	No	Yes	No	No	No	Yes	Yes	Yes	No*	Yes	5
Mahoney et al. (2005)	Yes	Yes	No	No	Unclear	Yes	Yes	Yes	Yes	Yes	7
Medrano et al. (2014)	No	No	No	No	Unclear	Yes	Yes	Yes	No*	Unclear	3
Ostojčić et al. (2014)	No	Unclear	No	Yes	Unclear	Yes	Yes	Yes	No	N/A	4
Sansoni et al. (2004)	No	Yes	No	No	No	Yes	Yes	Yes	No*	Unclear	4
Weaving et al. (2014)	No	Yes	No	Yes	Unclear	Yes	Yes	Yes	No*	Yes	6

Note. *Some factors/subgroups/differences identified and accounted for, but below level sufficient to rate as 'yes'; N/A = Not applicable; Item 1 = Sample representative of target population?; Item 2 = Appropriate recruitment method?; Item 3 = Adequate sample size?; Item 4 = Detailed description of participants and setting?; Item 5 = Data analysis conducted with sufficient coverage of sample?; Item 6 = Objective and standard measurement criteria used?; Item 7 = Reliable measurement used?; Item 8 = Appropriate statistical analysis?; Item 9 = Confounding factors/subgroups/differences identified and accounted for?; Item 10 = Subpopulations identified using objective criteria.

Synthesis of Anxiety Prevalence

Anxiety prevalence rates ranged from 3.7 percent (95% CI 1.4% to 9.4%, $p < 0.01$) to 76.5 percent (95% CI 59.5% to 87.8%, $p > 0.01$). In meta-analysis, a pooled prevalence estimate of 32.1 percent was achieved (95% CI 20.6% to 46.2%, $p = 0.01$). Visual inspection of the forest plot presented in Figure 2 suggested possible heterogeneity between studies, which was found to be statistically significant ($I^2 = 92.9\%$, $p < 0.01$).

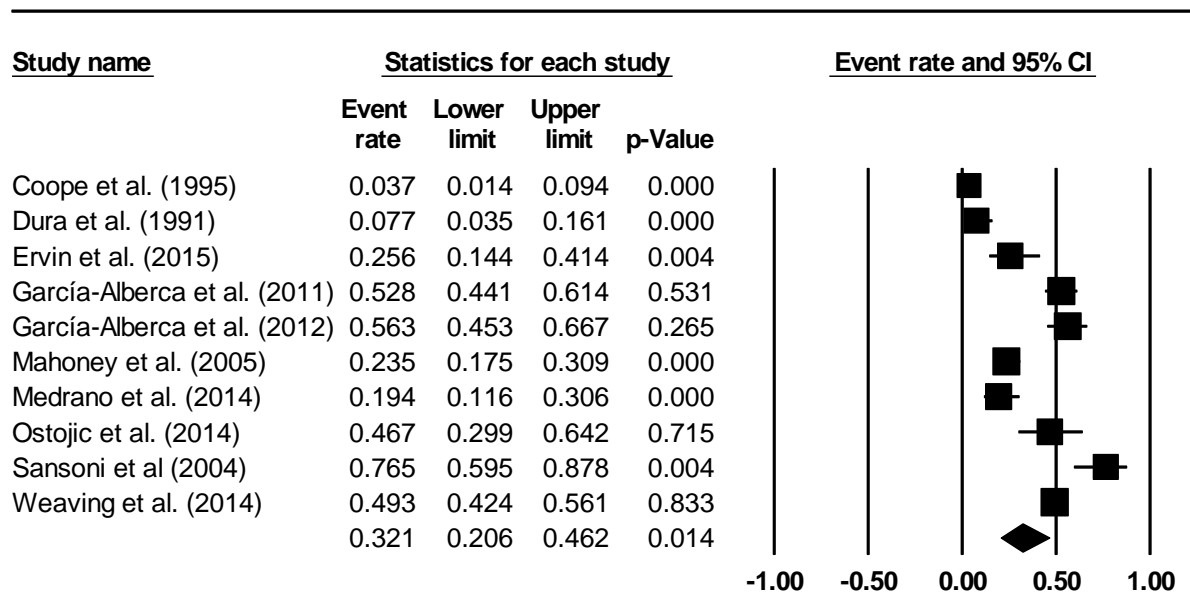


Figure 2. Forest plot for meta-analysis of anxiety prevalence

Publication bias

Visual inspection of the funnel plot for all included studies was inconclusive (Appendix D). Duval and Tweedie's (2000) trim and fill method imputed one missing study estimating an adjusted prevalence rate of 27.64 percent (95% CI 16.73% to 42.07%). Rosenthal's (1983) fail-safe N calculated 115 missing studies would be required to increase the two-tailed p-value to above .05.

Sensitivity analysis

Sansoni *et al.* (2004) met several of the criteria for exclusion in sensitivity analysis. Firstly, the study was identified on the forest plot as a possible outlier (Figure 2).

Secondly, the eligibility of Sansoni *et al.* (2004) contained some uncertainty due to the use of a higher than usual symptom cut-off score. Thirdly Sansoni *et al.* (2004) was rated as poor quality, achieving only three out of ten positive items. A further two studies were excluded in the sensitivity analysis based on low quality. Both Medrano *et al.* (2014) and Ostojić *et al.*, (2014) achieved only four out of ten positive items. The sensitivity analysis yielded a reduced though comparable pooled prevalence rate of 27.0 percent (95% CI 15.4% to 42.9%, $p<0.01$). Heterogeneity remained statistically significant ($I^2=94.0%$, $p<0.01$).

Subgroup analysis

Subgroup analysis was initially conducted using all included studies. A statistically significant difference in prevalence rate between studies grouped by prevalence measurement tool type was identified ($p<0.01$). The pooled prevalence rate as measured by a diagnostic tool was 5.6 percent (95% CI 2.7% to 11.3%, $p<0.01$), with no statistically significant heterogeneity between studies ($I^2=28.2%$, $p=0.24$). The pooled prevalence rate as measured by a self-report symptom measure was 42.6 percent (95% CI 30.96% to 55.3%, $p<0.25$), with statistically significant heterogeneity between studies ($I^2=89.6%$, $p<0.01$). The obtained prevalence rates were similar when the subgroup analysis was conducted after sensitivity exclusions (diagnostic tool=5.6% prevalence, 95% CI 2.7% to 11.3%, $p<0.01$; self-report symptom measure=41.2% prevalence, 95% CI 28.6% to 55.1%, $p<0.22$). Further subgroup analysis using individual self-report measures as subgroups was not conducted due to a large variety in the measures used and the small number of studies using each measure.

Subgroup analysis was not conducted using dementia type as there were no studies using a sample of caregivers for a single dementia type, apart from Alzheimer's dementia. Furthermore, subgroup analysis was not conducted on HDI category due to all but one study country being rated as 'very high'.

Discussion

Anxiety is arguably somewhat neglected in the dementia caregiver literature. It is therefore important to establish if anxiety is a prevalent difficulty experienced by dementia caregivers. This will help to ensure caregiver interventions are suitably tailored to reflect the difficulties experienced by dementia caregivers, in terms of both intervention approach/content, and outcome measurement. This study synthesized an estimate of the prevalence of anxiety caseness in the dementia caregiver population, defined as anxiety diagnoses or a clinically relevant level of symptoms.

Overall Prevalence of Anxiety

The adjusted prevalence rate following publication bias assessment will not be discussed as it has been suggested that publication bias assessment is not reliable when fewer than 30 studies are included (Cuijpers, 2016). The overall pooled anxiety prevalence rate was 32.1 percent. By comparison, a previous meta-analysis of the global prevalence of anxiety disorders in the general population yielded a prevalence rate of 7.3 percent (Baxter *et al.*, 2013). This suggests that anxiety is indeed a prevalent psychological difficulty experienced by informal caregivers for people with dementia.

The pooled prevalence rate contained significant heterogeneity across individual study prevalence rates. Excluding Sansoni *et al.* (2004), Medrano *et al.* (2014) and Ostojčić *et al.* (2014) in sensitivity analysis reduced the prevalence estimate to 27 percent but did not reduce heterogeneity. The individual prevalence rates for both Sansoni *et al.* (2004) and Ostojčić *et al.* (2014) were greater than the overall obtained prevalence rate, whilst Medrano *et al.* (2014) obtained a lower prevalence rate. The reduction in prevalence was somewhat unexpected given that Sansoni *et al.* (2004) used a higher than usual cut-off score to determine caseness, which was expected to result in an underestimation of prevalence. However, it was noted that Sansoni *et al.* (2004) was the only study to include a female only sample, and prior research has suggested that female caregivers experience greater fear and psychological morbidity compared to male caregivers

(Mausbach *et al.*, 2013). The female gender sample may therefore have resulted in a greater anxiety prevalence rate compared to mixed gender samples.

Diagnostic tools versus self-report symptom measures

Subgroup analysis revealed a statistically significant difference in prevalence as measured by diagnostic tools and self-report symptom measures. The pooled prevalence rate when a diagnostic tool was used was 5.6 percent, and whilst no significant heterogeneity was found, it is possible that the small number of studies in the diagnostic tool group ($n = 2$) did not allow for the detection of heterogeneity (Cuijpers, 2016). The self-report symptom measure prevalence was significantly higher at 42.6 percent, though the pooled estimate was not statistically significant which may be a reflection of the small number of included studies. Furthermore, significant heterogeneity remained. Further exploration of heterogeneity was not conducted as subgroup analysis was limited by the number and characteristics of included studies.

These results may appear to suggest that the different measurement tools were measuring different constructs, and that the prevalence of diagnosable anxiety disorders was lower than the prevalence of caseness level symptoms. However, the self-report symptom measures used by included studies have been shown to have appropriate diagnostic specificity and sensitivity (Julian, 2011; Maier *et al.*, 1988).

When examining the two studies included in the diagnostic tool group, it was noted that Coope *et al.* (1995) did not include caregivers for people with 'severe' dementia', and the number of hours spent caregiving was not reported, though caregivers only needed to be in contact with the care-recipient once per week to be included. It is possible that the Coope *et al.* (1995) participants engaged in less caregiving, for people with less severe dementia, and both factors have been postulated to impact on psychological difficulties in caregivers (Brodaty and Donkin, 2009; Gaugler *et al.*, 2003). Furthermore, the Dura *et al.* (1991) sample had the lowest number of caregiving hours compared to the other studies

which reported caregiving hours. Dura *et al.* (1991) had reported a positive correlational relationship between the number of hours caregiving and depression scores, but had neglected to examine anxiety, or to refer to anxiety at all in their discussion.

Therefore, given the small number of studies included in diagnostic tool subgroup, and the lack of statistical significance in the pooled prevalence rate for the self-report symptom measure subgroup, it would be unwise to draw any conclusions regarding the moderating effect of different measurement tools.

Care-recipient dementia type

The majority of studies recruited only caregivers who cared for a person with Alzheimer's dementia. A minority of studies recruited caregivers who cared for a person with any type of dementia. In these studies, it was noted that the majority of caregivers cared for a person with Alzheimer's dementia. Whilst this reflects the prevalence of different types of dementia, it did not allow for the moderating impact of care-recipient dementia type on anxiety prevalence to be explored in this meta-analysis (Walsh, no date). However, it is possible that prevalence of anxiety is greater or reduced in caregivers for people with different types of dementia, given the varying difficulties associated with different dementia illness (Johnson *et al.*, 2011).

Country development status

All of the studies included in this meta-analysis used samples from countries categorised as very highly developed, apart from Medrano *et al.* (2014) which used a sample from a highly developed country, based on the HDI category of each country (United Nations Development Programme, 2016). It was therefore not possible to examine the moderating impact of country development status. The lack of studies from less developed countries may reflect a lack of research in these areas, though it is important to acknowledge that language bias may be a contributing factor as only studies reported in English were included. Given that dementia is a global difficulty, and that there may be a

greater demand for informal care in less developed countries, it is important that dementia caregiver research does not neglect the caregiver population in less developed countries (Prince *et al.*, 2013).

Anxiety and depression prevalence

This review sought to determine a pooled estimate of anxiety prevalence only. The overall prevalence of anxiety was 32.1 percent which is comparable to the pooled estimate of 31.2 percent for the prevalence of depression obtained by Collins and Kishita (2018). This suggests anxiety should be afforded as much consideration as depression in the caregiver literature, in terms of developing an understanding of moderating and mediating factors, and also with regards to intervention development and assessment.

Neither this review nor Collins and Kishita (2018) examined anxiety and depression comorbidity. However, it has been reported that depression often occurs with comorbid anxiety in dementia caregivers (Mahoney *et al.*, 2005). Rumination has been proposed as a transdiagnostic mechanism mediating anxiety and depression in a non-caregiver population, and there is emerging evidence that it may also mediate anxiety and depression in dementia caregivers (Hsu *et al.*, 2015; Márquez-González *et al.*, 2015). However, the comorbidity of anxiety and depression in dementia caregivers requires further exploration. Understanding the trajectory towards comorbid anxiety and depression in dementia caregivers, and any mechanisms common to both difficulties may help target interventions so that they may be more effective and resource efficient.

Additional Clinical and Research Implications

This study has highlighted that anxiety is indeed a prevalent difficulty experienced by dementia caregivers. It is therefore necessary that services supporting caregivers for people with dementia offer assessment and support for anxiety. It is also important that future research does not neglect anxiety in the dementia caregiver population.

Specifically, factors which moderate anxiety prevalence require more exploration, given

the high heterogeneity found in this study. Indeed, a previous review highlighted a lack of conclusive evidence regarding factors associated with anxiety prevalence (Cooper *et al.*, 2007). The present study suggests there has been little development regarding understanding moderators of anxiety prevalence over the past ten years. Additional dementia caregiver anxiety prevalence studies may allow for future meta-analyses to examine potential moderators such as care-recipient dementia type and country development status. Doing so may help to both identify caregivers which may be more vulnerable to experiencing anxiety, and in achieve a better understanding of potential protective factors. Larger scale cross-sectional studies and longitudinal studies affording consideration to various potential moderators would likely be beneficial.

Furthermore, given that this study has demonstrated anxiety is a prevalent difficulty for dementia caregivers, it justifies the need to better understand the underlying mediating factors. It would likely be beneficial for future research to also consider common transdiagnostic factors mediating the range of difficulties experienced by dementia caregivers. Should such common factors be identified, it may allow for the development of interventions which are beneficial for a variety of dementia caregivers, regardless of their primary presenting difficulties.

Strengths and Limitations

A strength of this meta-analysis is that it provides an updated pooled estimate of anxiety in caregivers for all types of progressive dementia. The pooled prevalence rate obtained justifies focusing future research on understanding underlying factors which may mediate anxiety, and how interventions may be adapted to address such factors. It also justifies the inclusion of anxiety as an outcome measure in dementia caregiver intervention research.

This study aimed to estimate the pooled prevalence of anxiety only. However, obtaining a pooled estimate of anxiety and depression comorbidity would have been

beneficial, as it was noted that five of the studies included in this review were included in Collins and Kishita's (2018) meta-analysis of the prevalence of depression (Sansoni *et al.*, 2004; Mahoney *et al.*, 2005; García-Alberca *et al.*, 2012; Medrano *et al.*, 2014; Ostojić *et al.*, 2014). However, only one of these studies reported on anxiety and depression co-morbidity (Mahoney *et al.*, 2005).

An important limitation of this study is the small number of studies included. A total of ten studies were included, which was reduced to seven following exclusion of low quality studies in sensitivity analysis. However, the cut-off score used to identify a study as low quality could be considered somewhat arbitrary, and as such exclusions based on quality should be interpreted with caution.

Another limitation is the large amount of statistically significant heterogeneity which could not be explained. As such, the overall prevalence rate obtained should be interpreted with caution. It was not possible to conclude the influence of possible moderating factors on the obtained heterogeneity. This was due to the limited number of studies in each moderator subgroup. Therefore, we do not know which caregiver, care recipient, or environmental factors may protect against, or increase the risk of caregiver anxiety. Developing a better understanding of both risk factors and protective factors for anxiety in the dementia caregivers may influence the focus of interventions. Therefore, this highlights the need for more research into the prevalence of anxiety in dementia caregiver populations.

Conclusions

This meta-analysis demonstrates clinical anxiety is a prevalent difficulty for caregivers of people with dementia. It suggests that anxiety is as prevalent as depression in this population, and as such warrants a similar level of focus in the research literature. The importance of expanding research into the prevalence of anxiety in less typically researched groups of dementia caregivers (e.g. caregivers residing in less developed

countries, caregivers for people with dementia illnesses other than Alzheimer's dementia), and exploring additional moderating and mediating factors is highlighted. Furthermore, this study supports including anxiety as an outcome measure in intervention research.

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Chapter 3. Bridging Chapter

Neglected Anxiety in Dementia Caregivers

The previous chapter demonstrates anxiety is a prevalent difficulty for dementia caregivers. Conversely, it was found that relatively few studies had examined the prevalence of anxiety. This supports the hypothesis that anxiety is somewhat neglected in the dementia caregiver literature.

It is not apparent why such a neglect exists, however the literatures points to several hypotheses. Firstly, anxiety is also neglected in the general older adult population, both clinically and in epidemiological and intervention research (Laidlaw, 2015). It has been suggested that anxiety may be overlooked in the older adult population due to challenges in identifying anxiety in older adults (Laidlaw, 2015). Specifically, there is an overlap of the physical symptoms of anxiety with the symptoms of some physical health conditions which commonly occur in later life. This may impact on the detection of anxiety in older adults, where anxiety may be overlooked as a cause of such symptoms if a possible physical explanation also exists (Bryant, Jackson, & Ames, 2008). Given that the majority of dementia caregivers fall within the older adult age range, and dementia caregivers are at an increased risk of experiencing a deterioration in their physical health, it is possible that dementia caregiver anxiety is overlooked for similar reasons (Alzheimer's Society, 2015; Schulz & Sherwood, 2008).

Secondly, it could be hypothesised that there are factors which may contribute to dementia caregivers being less likely to seek support for anxiety. It has been suggested that attitudes and beliefs towards health vary based on age group, or age 'cohort' (Hibbard & Pope, 1985). It has also been noted that cohort beliefs about seeking help for mental health concerns tend to vary, and some older aged cohorts can hold negative beliefs about seeking support for psychological difficulties (Mackenzie, Scott, Mather, & Sareen, 2008; Segal, Coolidge, Mincic, & O'Riley, 2005). Given that avoidance is a common feature of anxiety, older aged dementia caregivers who experience anxiety but

also hold negative cohort beliefs about seeking help may be more likely to avoid accessing support (Laidlaw, 2013; Segal et al., 2005). This may contribute to the underestimation of the importance of anxiety in dementia caregivers in clinical settings.

Thirdly, anxiety in later life is sometimes reasoned as justifiable or understandable by professionals, as opposed to a psychological difficulty which may benefit from support (Laidlaw, 2015). This has been described as a 'fallacy of good reasons', and again can result in anxiety being overlooked in older adults (Laidlaw, 2013). Given the challenges associated with caring for a person with dementia, anxiety in dementia caregivers of all ages may be viewed as understandable and therefore not requiring of further exploration both clinically and in the research literature.

In addition, professionals who perceive clients' anxious thoughts as objective and realistic, are more likely to feel pessimistic about client outcomes (Charlesworth & Greenfield, 2004). This negative bias has been demonstrated towards older adults seeking psychological support, and can influence the therapeutic relationship and outcome (Charlesworth & Greenfield, 2004). Such a bias may also occur towards dementia caregivers experiencing anxiety, and may contribute to the neglect of anxiety in this population.

However, none of these hypotheses can fully explain why anxiety is neglected in dementia caregivers. It is important to remember that not all dementia caregivers are older aged adults, and much of the evidence supporting the above hypotheses refers specifically to that age group. It is likely that several factors may contribute to the neglect of anxiety in dementia caregivers, and further research is required to fully understand what these factors are.

Interventions for Anxiety in Dementia Caregivers

Concordant with the paucity of epidemiological research investigating anxiety in dementia caregivers, prior reviews have identified a lack of interventions which target and

measure anxiety as an outcome in the dementia caregiver population (Cooper, Balamurali, Selwood, & Livingston, 2007; Elvish, Lever, Johnstone, Cawley, & Keady, 2013). As such, interventions for dementia caregivers should consider addressing anxiety, and should include measures of anxiety when assessing intervention outcomes.

Cognitive-Behavioural Therapy

Cognitive-Behavioural Therapy (CBT) is a psychological intervention based on cognitive-behavioural theory, which has extensive empirical support for reducing anxiety and other psychological difficulties in a variety of populations (Hofmann, Asnaani, Vonk, Sawyer, & Fang, 2012).

Cognitive-behavioural theory stipulates that thoughts and perceptions can influence emotions and physical feelings, which then impact on behaviour (A. Beck, 1976). It is postulated that change in any one of these four areas (thoughts, emotions, physical feelings, behaviour) can influence the others (J. Beck, 2011). As such, environmental or internal events which are perceived in a distorted or unhelpful manner can evoke strong emotions (e.g. anxiety) and lead to maladaptive behaviours (J. Beck, 2011). Such maladaptive behaviours are often self-reinforcing in that the behaviour (e.g. avoidance) leads to an immediate reduction in the strong emotion (e.g. anxiety), which is reinforced via operant conditioning mechanisms (Skinner, 1953). However, such behaviours can have longer-term negative consequences which can perpetuate a cycle of distress (J. Beck, 2011).

For example, a caregiver who is asked about their plans for the next year, may have thoughts that they will not be able to cope with supporting their family member who has dementia, and then feel strong anxiety. They may subsequently avoid experiencing the emotion by, for example, avoiding social situations where such conversations arise. This may reduce the feelings of anxiety in the short-term. However, avoiding social situations may inadvertently increase anxiety about such situations, and may also reduce

social support from other friends and family members. This may strengthen their thoughts that they cannot cope, increasing feelings of anxiety, and increasing the need to engage in maladaptive coping strategies.

CBT is based on the cognitive-behavioural theory described above and is the most recommended psychological therapy in NICE guidelines for various mental health diagnoses (NICE, 2009; NICE, 2011). CBT for anxiety often involves identifying and altering unhelpful cognitive appraisals, and behavioural exposure-based techniques (Kaczurkin & Foa, 2015). Meta-analytic evidence supports the use of CBT compared to control interventions for various anxiety disorders (e.g. generalised anxiety disorder, obsessive-compulsive disorder, post-traumatic stress disorder) (Carpenter et al., 2018; Hofmann & Smits, 2008). CBT is also supported for anxiety in both an adult and an older adult population (Gould, Coulson, & Howard, 2012). These are age groups which are often caregivers for people with dementia, usually as spouses or adult children (Alzheimer's Society, 2014).

The literature examining CBT for anxiety in dementia caregivers is somewhat limited. Research into CBT for dementia caregivers tends to focus on depression as an outcome (Kwon, Ahn, Kim, & Park, 2017). Nonetheless, there are reports that CBT has a significant effect on reducing anxiety outcomes in dementia caregivers (Cooper, Balamurali, Selwood, et al., 2007; Kwon et al., 2017). However, it is important to note that a recent meta-analytical review reported that CBT did not produce a statistically significant pooled effect on anxiety scores in dementia caregivers (Hopkinson, Reavell, Lane, & Mallikarjun, 2018). Whilst this appears to contradict the previous statement, it is important to note that only one of the ten included studies tailored the intervention to anxiety as a primary outcome, and in that study a significant decrease in anxiety was obtained (Akkerman & Ostwald, 2004).

Therefore, based on the above, the literature regarding the use of CBT for anxiety in dementia caregivers can be described as promising, but limited in amount. It is clear

that further research is required, particularly research that focuses specifically on the use of CBT for anxiety in dementia caregivers.

Additional Dementia Caregiver Outcomes

The first part of this thesis portfolio focuses on anxiety in dementia caregivers. However, depression is arguably the most commonly reported psychological difficulty in the literature, and commonly a main outcome in dementia caregiver intervention research (Collins & Kishita, 2018; Cuijpers, 2005; Kwon et al., 2017; Schulz et al., 1995). Burden and perceived stress or strain have also been identified as important psychological wellbeing variables (Pinquart & Sörensen, 2003; Schulz et al., 2002). Furthermore, it has been recommended that any studies that examine caregiver interventions should include a core set of outcomes addressing clinical symptomology (e.g. anxiety; depression), quality of life (e.g. psychological wellbeing; life satisfaction), social validity (e.g. the acceptability of an intervention) and social significance (e.g. placement of care recipient in formal residential care) (Schulz et al., 2002). The consistent use of such outcomes is expected to aid the ability to draw meaningful conclusions from the diverse range of dementia caregiver interventions and outcomes in the literature (Schulz et al., 2002). Whilst these recommendations were presented 15 years previously, and it has been noted that few studies examine all of the above areas of outcome (likely due to pragmatic limitations), most dementia caregiver intervention research studies focus on an area of outcome previously recommended by Schulz et al. (2002), suggesting continued relevance to the dementia caregiver population and associated stakeholders (e.g. service providers) (Boots, De Vugt, Van Knippenberg, Kempen, & Verhey, 2014; Roche, Croot, MacCann, Cramer, & Diehl-Schmid, 2015; Tanner et al., 2015).

Second Empirical Paper

The empirical paper presented in the next chapter synthesizes the literature on low-intensity CBT-based interventions. The study aims to obtain an empirically derived

index of the efficacy of low-intensity CBT on psychologically relevant outcomes among dementia caregivers. The outcomes are selected based both on chapter two of this portfolio, and the above mentioned recommendations of Schulz et al. (2002). As such, the empirical paper includes examines the outcomes of anxiety and depression as measures of clinical symptomology, and burden and perceived stress/strain (labelled as 'distress') as psychological wellbeing quality of life measures. Social validity and social significance are taken into consideration in a study quality assessment, but not reported on as outcomes as the primary aim is to focus of psychologically relevant outcomes.

The rationale for quantitatively synthesizing the literature on low-intensity CBT for dementia caregivers is provided within the empirical paper.

Chapter 4. Empirical Paper Two

This chapter consists of a systematic review and meta-analysis, written for publication in the journal *International Psychogeriatrics*. This chapter is formatted in accordance with the journal submission guidelines (Appendix A).

Abstract word count: 208 (250 words journal limit)

Total word count (excluding abstract, tables, figures and appendices): 4888 (5000 words journal limit)

Running Head: LOW-INTENSITY CBT FOR DEMENTIA CAREGIVERS

A meta-analysis of low-intensity cognitive behavioural therapy-based interventions for
dementia caregivers

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Abstract

Objectives: This study aimed to review the effectiveness of low-intensity cognitive behavioural therapy (CBT)-based interventions for informal dementia caregivers, when compared to non-active control conditions.

Design: Literature searches were conducted in databases of published (PsycINFO; MEDLINE; CINAHL; Scopus) and unpublished (Open Grey; ISRCTN registry; ClinicalTrials.gov; ProQuest) literature. Individual meta-analyses were conducted for each outcome variable. Pooled intervention effect estimates were calculated as Hedge's g using a random-effects model.

Included studies: Studies examining the effect of low-intensity CBT-based interventions for informal caregivers for people with any progressive were included. Randomised-controlled trials and controlled clinical trials were included.

Measurements: Outcomes included the psychological variables of anxiety, depression, burden and distress (defined as stress or strain).

Results: A total of five studies reported anxiety outcomes, twelve reported on depression, three reported on burden and six reported distress outcomes. Results demonstrated a significant effect of low-intensity CBT-based interventions in reducing all examined psychological difficulties. Small effects sizes were found for anxiety ($g = 0.35$), depression ($g = 0.27$) and distress ($g = 0.33$). A medium effect was found for burden ($g = 0.53$).

Conclusions: The results provide initial support for low-intensity CBT-based interventions for dementia caregivers. Clinical implications and research recommendations are explored. Strengths and limitations of the study are discussed.

Keywords

Dementia caregivers; cognitive behavioural therapy; CBT; low-intensity; anxiety; depression; burden; distress

Introduction

Dementia illnesses have a degenerative impact on the neurocognitive abilities of those affected (Knapp *et al.*, 2007). Dementia is associated with significant behavioural changes and is both progressive and incurable (McKeith and Cummings, 2005). As such, the care needs for people with dementia generally increase as the disease progresses (Knapp *et al.*, 2007).

Many people with dementia are cared for by informal caregivers who are often family members (Friedman *et al.*, 2015). Caring for a person with dementia can have a negative impact on the carer's physical, financial, social and psychological wellbeing (Brodaty and Donkin, 2009; Bennett *et al.*, 2013). Dementia caregivers (DC) are more likely to experience burden, depression, anxiety and stress compared to both the general population and caregivers for people with other illnesses (Schulz *et al.*, 1995; Bertrand *et al.*, 2006). Furthermore, psychosocial difficulties in DCs have been associated with an increase in care recipients being placed in formal residential care (Brodaty and Donkin, 2009).

This is important given that the global prevalence of dementia was estimated at 35.6 million people in 2010, with a prediction that this number will double every 20 years (Prince *et al.*, 2013). As such it is expected that there will be an increase in the need for informal care. The need for informal care may be greater in less developed countries, where access to formal care is more limited, and in countries with chronically underfunded health and social care systems (Prince *et al.*, 2013; Franca, 2017).

Concordantly, the need to develop effective, low-cost, and accessible interventions for DCs has been recognised by the World Health Organisation (World Health Organisation, 2012). Currently, a variety of DC interventions exist, addressing the wide range of difficulties associated with caregiving. Such interventions include

providing education about dementia and caregiving, respite care, general support, psychological interventions and multi-component interventions which may combine aspects of several types of intervention (Pinquart and Sörensen, 2006).

Previous comprehensive reviews of DC interventions suggested that psychoeducational programmes and Cognitive-Behavioural Therapy (CBT) can impact on the wellbeing of DCs, with the latter approach shown to be more effective for psychological difficulties (Pinquart and Sörensen, 2006; Gallagher-Thompson and Coon, 2007; Elvish *et al.*, 2013). CBT is a psychological intervention with growing global empirical support for DCs, particularly with regards to depression outcomes (Kwon *et al.*, 2017). It has been suggested that CBT alters negative caregiving related appraisals, reduces the use of unhelpful coping strategies and encourages caregivers to engage in positive activities (Laidlaw, 2015).

However, CBT can be a resource-intensive intervention. Specific higher-education qualifications are usually required to license a therapist to deliver CBT, though the exact requirements are dependent upon the country in which it is being delivered (BABCP, 2012; NACBT, 2016). A recent review conducted by Kwon *et al.* (2017) reported the number of CBT sessions delivered for DCs ranged from 8 to 13 sessions, and such a number of sessions could be described as resource intense when delivered by accredited/licensed therapists. Less developed and poorly funded countries have less access to licensed health professionals, and are therefore less able to provide resource-intensive psychological interventions such as high-intensity CBT (World Health Organization, 2014). Conversely, these are the areas which are likely to experience a greater demand for informal care. However, it remains yet to be determined if 'lower-intensity' CBT interventions are effective in reducing the psychological difficulties associated with dementia caregiving.

Low-intensity CBT interventions are those which are based on Cognitive-Behavioural theory and are either not facilitated (e.g. CBT self-help), are facilitated by

non-highly qualified facilitators, or are facilitated by highly qualified facilitators for only a short duration (Bennett-Levy *et al.*, 2010). Low-intensity CBT-based interventions have gained support in the United Kingdom as part of a 'stepped-care' model of public health (NICE, 2011). The stepped-care model stipulates that the most effective, yet least resource intensive intervention should first be delivered before 'stepping-up' to a higher level of intensity (Bennett-Levy *et al.*, 2010). It not clear if adopting such an approach is beneficial to DCs experiencing psychological difficulties as previous reviews of CBT for DCs do not consider the intensity of interventions.

Given the above mentioned limitations in resources, it is important to assess the effectiveness of low-intensity CBT for DCs on a range of outcomes relevant to DCs (Schulz, *et al.*, 2002). Therefore, this study aims to examine the effectiveness of low-intensity CBT-based interventions for DCs on four primary outcomes of anxiety, depression, burden, and distress (defined as perceived stress or strain).

Method

Protocol and Registration

The review protocol was published on the PROSPERO international prospective register of systematic reviews (registration number: CRD42017060105; accessed via www.crd.york.ac.uk/PROSPERO).

Search Strategy

A systematic search of published literature was conducted using the electronic databases PsycINFO, MEDLINE, CINAHL and Scopus. A search of unpublished literature was conducted to address potential publication bias, using Open Grey, ProQuest, the ISRCTN registry and ClinicalTrials.gov. Reference lists of key review papers were hand searched. Key search terms included (a) dementia, (b) caregivers, (c) CBT-based therapy, (d) clinical trials, and related terms. Terms and limits were adapted to each

source (Appendix E). Sources were searched from the date of database inception to July 1st 2017.

Eligibility Criteria

Articles were eligible if the following criteria were met: (a) Participants were informal adult caregivers for a person with an organic dementia illness. (b) The intervention was based on CBT theory or techniques (A. Beck, 1979), The intervention may include techniques that encourage caregivers to target their situation, thoughts, feelings (physical/emotional) or behaviour. For example, problem solving, thought challenging, behavioural activation, graded exposure etc. (J. Beck, 2011). Eligible CBT-based interventions included CBT psychoeducation, CBT therapy, multicomponent CBT-based interventions and third-wave CBT interventions, such as Acceptance and Commitment Therapy. (c) The intervention was low-intensity, as based on a public health stepped-care model for anxiety and depression, and existing review literature on low-intensity interventions, due to a lack of a universal definition (NICE, 2009, 2011a, 2011b; Richards *et al.*, 2010; Rodgers *et al.*, 2012). This was defined as interventions delivered by facilitators locally licensed/accredited to practice CBT independently, lasting no more than six hours; interventions delivered by facilitators who are not licensed/accredited to practice CBT independently, lasting any duration; and self-help interventions. Interventions with mixed facilitators lasting more than six hours were included if the accredited/licensed facilitation did not exceed 6 hours. (d) The study reported outcomes for at least one of the four outcomes of interest: anxiety, depression, burden and distress. Distress was defined as perceived stress or strain, as described by Cohen (1983), and burden followed the description provided by Zarit (1980). (e) The study was a randomized controlled trial (RCT) or controlled clinical trial (CCT) using a waitlist, treatment as usual, placebo, or non-active intervention comparison group. Non-active interventions were defined as: (i) Interventions which are expected to have no effect on the outcome variables, based on prior research or theory. (ii) Interventions designed to be similar to treatment as usual. (iii)

Interventions which match the treatment intervention in all aspects except for the expected CBT based active component, which must be replaced by a known non-active component (Karlsson and Bergmark, 2015).

Study Selection

The primary reviewer (LK) identified potentially relevant articles found in the search based on the abstracts. Duplicates were excluded and full reports of the remaining articles were obtained and assessed for eligibility by LK and the secondary reviewer (NK). Discrepancies were discussed and resolved, and a third reviewer was available for consultation if unresolved. Additional information was requested from authors of 27 articles where information was insufficient to determine eligibility, via a contact procedure (Appendix F). Missing information related to intervention intensity and frequently included facilitator profession/qualification level. Articles were subsequently excluded if additional information was not provided (Figure 3).

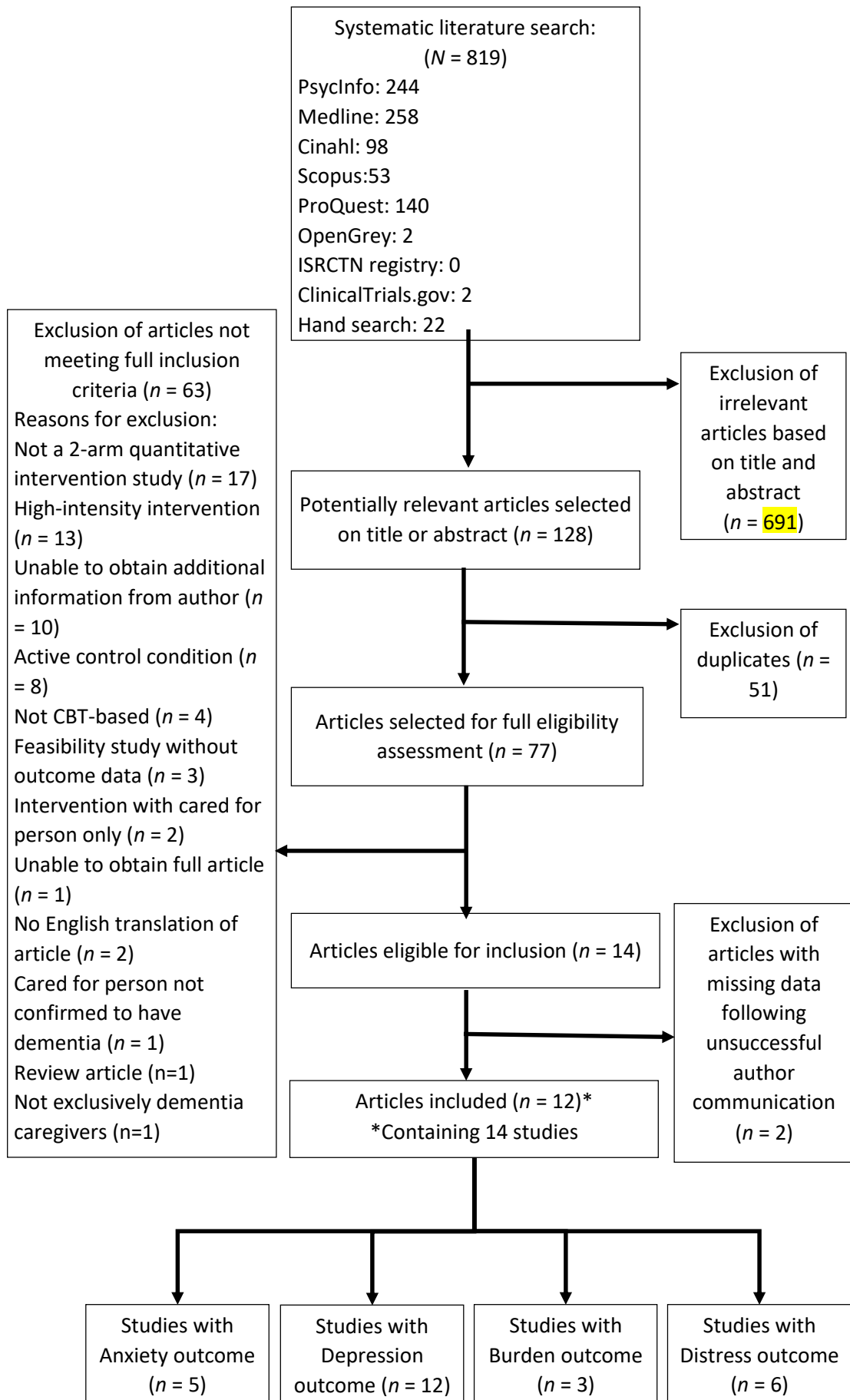


Figure 3. Systematic literature search flow chart

Quality Assessment & Risk of Bias

The quality and risk of bias of eligible studies was assessed using the RCT Psychotherapy Quality Rating Scale (RCT PQRS) which was adapted to reflect important qualities for low-intensity DC intervention RCTs (Kocsis *et al.*, 2010). The scale rates the quality of studies based on the description of participants, definition and delivery of the intervention, outcome measures, data analysis, intervention assignment and overall study quality. Consideration of selection, detection, and attrition bias are included within these areas. The adapted RCT PQRS was a 27 item electronic spreadsheet scale (Appendix G), Items relating to facilitation were omitted for self-help interventions. The scale included the addition of participant items relating to both the caregiver and cared for person, and an additional outcome item assessing the inclusion of measures of social acceptability and social validity as recommended by Schulz *et al.* (2002). The completed scale provided a quality percentage score and one of seven qualitative classification descriptors ranging from exceptionally poor to exceptionally good. Studies which were rated as 'very poor' (16 to 29%) or 'exceptionally poor' ($\leq 15\%$) were excluded. The adapted RCT PQRS was piloted by LK and NK. The scale was then completed independently for all included studies by LK and an additional reviewer (AS), and any discrepancies were discussed and resolved. Unresolved discrepancies were discussed with the third reviewer (NK) and resolved.

Data Extraction

An electronic data extraction form (Appendix H) was used to extract study characteristics, participant information, intervention and control group characteristics and continuous outcome data, as per Cochrane guidelines (Higgins and Green, 2011). The data extraction form was piloted by LK and NK. Data was then extracted independently by LK and AS, and any discrepancies were discussed and referred to NK if not resolved. Missing outcome data was sought from authors and studies were excluded from quantitative analysis if data was not obtained. Five studies required additional outcome

data from authors, and two were subsequently excluded due to lack of communication (Figure 3).

Statistical Analysis

Statistical analyses were performed using Comprehensive Meta-Analysis software (Biostat Inc., 2014). The analyses used pre and post means (M), standard deviations (SD), and participant numbers (N) for intervention and control groups for each study. Data from studies with multiple eligible intervention conditions were treated as individual studies when independent control groups were used (Borenstein *et al.*, 2009). Studies containing multiple eligible intervention conditions that were compared to a single control group were merged to create a composite study to address statistical dependence (Scammacca *et al.*, 2014). Composite data was derived by calculating an overall M across intervention conditions, and a composite SD was determined using a reverse analysis of variance method (Borenstein *et al.*, 2009).

Separate meta-analyses were conducted for each of the four primary outcome variables. The Standardised Mean Difference (SMD) was calculated as Hedges' *g* for study outcomes to allow for the use of different instruments across studies (Higgins and Green, 2011). The pooled intervention effect estimate was calculated using a random-effects model, as this is most appropriate model when there is expected variation in intervention characteristics (Borenstein *et al.*, 2009; Higgins and Green, 2011; Cuijpers, 2016).

Heterogeneity was examined visually using a forest plot and outliers removed in sensitivity analysis. The I^2 statistic was calculated, which shows the percentage of the total variance which can be explained by heterogeneity (Cuijpers, 2016). Where there was evidence of heterogeneity of the treatment effect, subgroup analyses were conducted using intervention facilitation type (e.g. self-help, facilitated), delivery format (e.g. group, individual), delivery method (e.g. computerised, face-to-face), and intervention approach

(e.g. CBT psychoeducation, CBT multi-component) as moderators. A random effects meta-regression was planned to explore facilitator contact hours as a predictor of effect size.

Additional sensitivity analysis was conducted by excluding studies which appeared to meet the inclusion criteria but somewhat arbitrary or unclear compared to other included studies. Publication bias was explored visually using a funnel plot, and Orwin's Fail-safe N was calculated (Orwin, 1983; Borenstein *et al.*, 2009). The 'trim and fill' method was applied to estimate effects sizes after bias had been taken into account (Duval *et al.*, 2000).

Results

Study Selection

The search resulted in 12 eligible articles, with one article containing two eligible studies (Gallagher-Thompson *et al.*, 2008), and another containing two eligible interventions using the same control group (Steffen, 2000). This resulted in a total of 14 eligible studies as presented in Figure 3. For the purposes of data analysis, the studies within Gallagher-Thompson *et al.* (2008) were treated as individual studies (Gallagher-Thompson *et al.*, 2008a; 2008b). The intervention groups contained within Steffen (2000) were treated as individual studies for descriptive purposes (Steffen, 2000a; 2000b), however outcome data from both intervention groups was merged to form composite scores for the purposes of quantitative analysis, due the use of a single control group (Borenstein *et al.*, 2009).

Study Characteristics

Participant characteristics are summarised in Table 4. A total of 1131 participants were included in the analysis (602 intervention, 529 control), with mean ages ranging from 46.9 to 65.5 years. The majority were female spousal caregivers for people with either Alzheimer's disease or a non-specified dementia. All samples were from developed

countries, based on the Human Development Index (United Nations Development Programme, 2016).

Intervention characteristics are summarised in Table 5. The majority of studies delivered multi-component CBT-based interventions, combining CBT techniques with other non-CBT techniques (Chang, 1999; Beauchamp *et al.*, 2005; Gallagher-Thompson *et al.*, 2008a; 2008b; Tremont *et al.*, 2008; Au *et al.*, 2010; Villareal-Reyna *et al.*, 2012; Dowling *et al.*, 2013; Kajiyama *et al.*, 2013; Blom *et al.*, 2015; Steffen and Gant, 2016). A minority of studies delivered interventions using only CBT-based techniques (Steffen, 2000a; 2000b; Chiu *et al.* 2015). There were no passive CBT-based psychoeducation only interventions, as all interventions contained active components, and no third-wave CBT-based interventions. One study identified as a CBT-based multicomponent intervention was selected for removal in sensitivity analysis due a minority of CBT-based components identified within the intervention (Dowling, *et al.*, 2013).

Nine studies delivered individual interventions (Chang 1999; Steffen, 2000a; Beauchamp *et al.*, 2005; Tremont *et al.*, 2008; Dowling, *et al.*, 2013; Kajiyama *et al.*, 2013; Blom *et al.*, 2015; Chui *et al.*, 2015; Steffen and Grant, 2016) and five used a group method (Steffen, 2000b; Gallagher-Thompson *et al.*, 2008a; 2008b; Au *et al.*, 2010; Villareal-Reyna *et al.*, 2012).

Delivery methods were wide ranging and included face-to face (Gallagher-Thompson *et al.* 2008a; 2008b; Villareal-Reyna *et al.*, 2012; Au *et al.*, 2015; Chui *et al.* 2015), face-to-face and video (Steffen, 2000a), face-to-face and videoconference (Dowling *et al.*, 2013), telephone (Tremont *et al.*, 2008), telephone and video (Chang, 1999; Steffen, 2000b), telephone, video and bibliotherapy (Steffen and Grant, 2016), computerised (Beauchamp *et al.*, 2005; Blom *et al.*, 2015), and computerised and bibliotherapy (Kajiyama *et al.*, 2013). Nine studies were facilitated by non-CBT licensed facilitators (Chang, 1999; Steffen 2000a; 2000b; Gallagher-Thompson *et al.* 2008a; 2008b; Tremont *et al.*, 2008; Au *et al.*, 2012; Villareal-Reyna *et al.*, 2012; Chui, 2015), two

used mixed licensed and non-licensed facilitators (Dowling *et al.*, 2013; Steffen and Grant, 2016), one used licensed facilitators (Blom *et al.*, 2015) and two studies were non-facilitated self-help interventions (Beauchamp *et al.*, 2005; Kajiyama *et al.* 2013). The total facilitator contact time ranged from 0 to 26 hours.

Control groups included non-active interventions (Chang, 1999; Gallagher-Thompson *et al.* 2008a; 2008b; Villareal-Reyna *et al.*, 2012; Dowling *et al.*, 2013; Kajiyama *et al.*, 2013; Blom *et al.*, 2015), waitlist controls (Steffen, 2000a; 2000b; Beauchamp *et al.*, 2005; Au *et al.* 2010), and treatment as usual (Tremont *et al.*, 2008; Chui *et al.*, 2015; Steffen and Gant, 2016). Control and intervention group content overviews are provided in Appendix I.

Of the 14 studies, five included anxiety outcomes (Chang, 1999; Beauchamp *et al.*, 2005; Villareal-Reyna *et al.*, 2012; Blom *et al.*, 2015; Steffen and Grant, 2016), 12 included depression outcomes (Chang, 1999; Steffen, 2000a; 2000b; Beauchamp *et al.*, 2005; Gallagher-Thompson *et al.* 2008a; 2008b; Tremont *et al.*, 2008; Au *et al.*, 2012; Dowling *et al.*, 2013; Kajiyama *et al.*, 2013; Blom *et al.*, 2015; Steffen and Grant, 2016) three included burden outcomes (Tremont *et al.*, 2008; Dowling *et al.*, 2013; Chiu *et al.*, 2015) and six included distress as an outcome (Beauchamp *et al.*, 2005; Gallagher-Thompson *et al.*, 2008a; 2008b; Dowling *et al.*, 2013; Kajiyama *et al.*, 2013; Chiu *et al.*, 2015).

Table 4. Participant characteristics and quality of studies included in the meta-analyses

Study	Country	N	Mean age (SD)	Ethnicity (%)	% female	Relationship to care recipient (%)	Care recipient dementia type (%)	Study quality (% score)
Au et al. (2010)	China	27	NR	Chinese Cantonese (100)	100	Spouse (NR); Adult child (NR)	Alzheimer's	Moderately poor (40.7)
Beauchamp et al. (2005)	USA	299	46.9 (12.2)	Caucasian (88); African-American (4); Hispanic (8); Other (8)	73	Adult child (67); Other relative (23); Spouse (7); Non- relative (3)	Any dementia	Moderately poor (34.7)
Blom et al. (2015)	Netherlands	245	61.2 (12.37)	Dutch (99.2)	69.4	Spouse (58.4); Adult child (39.6)	Alzheimer's (74.5); Any dementia	Average (57.6)
Chang (1999)	USA	65	66.5 (NR)	Caucasian (79.1); African-American (16.3)	100	Spouse (NR); Adult child (NR)	Any dementia	Moderately poor (33.9)
Chiu et al. (2015)	Canada	54	NR	NR	83	Spouse (NR); Adult child (NR)	Any dementia	Average (50.8)

Dowling et al (2013)	USA	24	59.5 (8.1)	White (90), Hispanic & African-American (10)	70.8	Spouse (87.5); Other (NR)	Fronto-temporal	Moderately poor (35.6)
Gallagher-Thompson et al. (2008a)	USA	95	63.4 (13.66)	White (100)	100	Spouse (NR); Adult child (NR)	Any dementia	Average (54.2)
Gallagher-Thompson et al. (2008b)	USA	89	51.45 (11.55)	Hispanic/Latina (100)	100	Spouse (NR); Adult child (NR)	Any dementia	Average (54.2)
Kajiyama et al. (2013)	USA	103	NR	Caucasian (85); African-American (2.9); Asian-American (4.8); Hispanic-American (2.9); Native-American (1.9); Hawaiian/Pacific Islanders (0.9)	83	Spouse (NR); Adult child (NR); Other (NR)	Any dementia	Moderately good (63.3)
Steffen (2000)*	USA	28	64.06 (11.5)	Caucasian (75.8); African-American (24.2)	75.8	Spouse (54.5); Adult child (36.4); Other (9.1)	Alzheimer's (84.8); Other dementia (15.2)	Average (45.8)

Steffen and Grant (2016)	USA	46	60.3 (10.8)	White (79.7); African-American (20.3)	100	Spouse (52); Adult child (48)	Alzheimer's (62.2); Vascular (8.1); Lewy-body (2.7); Other dementia	Moderately good (62.7)
Tremont et al. (2008)	USA	33	NR	NR	NR	Spouse (33); Adult child (21.7)	Any dementia	Average (55.9)
Villareal-Reyna et al. (2012)	Mexico	23	NR	NR	NR	Adult child (NR); Non-specified family member (NR)	Alzheimer's	Average (54.2)

Note. *Composite study combining Steffen 2000a & 2000b; **Steffen 2000a; ***Steffen 2000b; N = Number of participants; NR = Not reported.

Table 5. Characteristics of interventions and outcome measures for each study

Study	Approach	Delivery format	Delivery method	Facilitation type	Total facilitator contact time (hours)	Outcome instruments	Measurement time points
Au et al. (2010)	CBT-based MC	Group	Face-to-face x13 2hr sessions	Guided by non-CBT licensed facilitators (trainee clinical psychologists)	26	CES-D	Pre; post
Beauchamp et al. (2005)	CBT-based MC	Individual	Computerised programme with no fixed duration, accessed 'as and when'	Self-help	0	BDI; STAI-S; CSI	Pre; post
Blom et al. (2015)	CBT-based MC	Individual	Computerised x8 sessions with email feedback	Guided by CBT licensed facilitator (psychologist)	NR	CES-D; HADS-A	Pre; post
Chang (1999)	CBT-based MC	Individual	Telephone & video x8 sessions of varied duration	Guided by non-CBT licensed facilitators (nurses)	NR	BSI-A; BSI-D	Pre; post; 1-month follow-up

Chiu et al. (2015)	CBT-based only	Individual	Face-to-face x3 1hr sessions	Guided by non-CBT licensed facilitators (care co-ordinators with a background in nursing, social work, occupational therapy or physiotherapy)	3	ZBI; PSS	Pre; post
Dowling et al (2013)	CBT-based MC	Individual	Face-to-face & videoconference x5 1hr sessions	Guided by mixed non-CBT licensed & licensed facilitators (nurse specialists and a psychologist)	5	CES-D; ZBI; PSS	Pre; post; 1-month follow-up
Gallagher-Thompson et al. (2008a)	CBT-based MC	Group	Face-to-face x12 2hr sessions	Guided by non-CBT licensed facilitators (psychology graduates and research fellows)	26	CES-D; PSS	Pre; post; 2-month follow-up
Gallagher-Thompson et al. (2008b)	CBT-based MC	Group	Face-to-face x12 2hr sessions	Guided by non-CBT licensed facilitators (psychology graduates and research fellows)	26	CES-D; PSS	Pre, post; 2-month follow-up
Kajiyama et al. (2013)	CBT-based MC	Individual	Computerised & bibliotherapy x6 modules with no time constraints	Self-help	0	CES-D; PSS	Pre; post

Steffen (2000)*	CBT-based only	Individual** Group***	Telephone & video x8 30min video & 20min phone call.** Face-to-face & video x8 90min group including a 30min video within each session***	Guided by non-CBT licensed facilitators (trainee clinical psychologists)	2.67	BDI	Pre; post
Steffen and Grant (2016)	CBT-based MC	Individual	Telephone, video & bibliotherapy x10 30 to 50min phone call & x10 30min video with workbook	Guided by mixed non- CBT licensed & licensed facilitators (x1 clinical psychologist, x5 non-licensed trainee psychologists)	NR	BDI-II; MAACL-R-A	Pre; post; 6- month follow- up
Tremont et al. (2008)	CBT-based MC	Individual	Telephone x23 phone calls, with initial calls lasting 1hr & follow-up calls lasting 15 to 30mins	Guided by non-CBT licensed facilitators (counsellor & psychology doctoral student)	12	GDS; ZBI	Pre; post
Villareal-Reyna et al. (2012)	CBT-based MC	Group	Face-to-face x8 90min sessions	Guided by non-CBT licensed facilitators (nurses)	12	ISA	Pre; post; 1- month follow- up

Note. *Composite study combining Steffen 2000a & 2000b; **Steffen 2000a; ***Steffen 2000b; NR = Not reported; CBT = Cognitive-Behavioural Therapy; MC = Multicomponent; CES-D = Center for Epidemiological Studies Depression Scale; BDI = Beck Depression Inventory; STAI-S = State-Trait Anxiety Inventory – State subscale; CSI = Caregiver Strain Instrument; HADS-A = Hospital Anxiety and

Depression Scale – Anxiety subscale; BSI-A = Brief Symptom Inventory – Anxiety subscale; BSI-D = Brief Symptom Inventory – Depression subscale; ZBI = Zarit Burden Interview; PSS = Perceived Stress Scale; MAACL-R-A = Multiple Affect Adjective Check-list Revised – Anxiety subscale; GDS = Geriatric Depression Scale; ISA = Inventory of State Anxiety.

Study Quality and Risk of Bias

The quality of included studies presented in Table 4 ranged from ‘moderately poor’ to ‘moderately good’. There were few large scale RCTs and the majority of studies were pilot or feasibility RCTs (Steffen, 2000a; 2000b; Tremont *et al.*, 2008; Au *et al.*, 2012; Villareal-Reyna *et al.*, 2012; Dowling *et al.*, 2013; Chiu *et al.*, 2015; Steffen and Grant 2016). Furthermore, few studies reported follow-up data, therefore it was only possible to calculate pre-post effects sizes, and not follow-up effects sizes.

Effect of Low-intensity CBT-based Interventions on Anxiety

Five studies evaluated the effect of a low-intensity CBT-based intervention on anxiety (Chang 1999; Beauchamp *et al.*, 2005; Villareal-Reyna *et al.*, 2012; Blom *et al.*, 2015; Steffen and Grant, 2016). All studies used differing tools to measure anxiety, including the state subscale of the State-Trait Anxiety Inventory (STAI-S) (Spielberger *et al.*, 1970; Beauchamp *et al.*, 2005), the anxiety subscales of the Hospital Anxiety and Depression Scale (HADS-A) (Zigmond and Snaith, 1983; Blom *et al.*, 2015), the Brief Symptom Inventory (BSI) (Derogatis, 1993; Chang, 1999) and the Multiple Affect Adjective Check-list Revised (MAACL-R-A) (Lubin and Zuckerman, 1985; Steffen and Gant, 2016), and the Inventory of State Anxiety (ISA) (Spielberger and Diaz-Guerrero, 2002; Villareal-Reyna *et al.*, 2012).

The individual study effects sizes as Hedge’s g ranged from 0.25 (95% CI -0.23 to 0.73, $p=0.31$) to 2.74 (95% CI 1.62 to 3.86, $p<0.01$). In meta-analysis, there was a significant reduction of anxiety yielding a pooled effect size of 0.58 (95% CI 0.17 to 0.97, $p<0.01$) (Appendix K). Statistically significant heterogeneity was found between studies ($I^2=78.82\%$ $p<0.01$), and examination of the forest plot identified Villareal-Reyna *et al.* (2012) as an outlier. Exclusion of Villareal-Reyna *et al.* (2012) yielded a pooled effect size of 0.35 (95% CI 0.20 to 0.50, $p<0.01$), with no statistically significant heterogeneity ($I^2=0.00\%$ $p<0.62$). Visual inspection of the funnel plot did not reveal publication bias

(Appendix L). Duval and Tweedie's (2000) trim and fill method did not impute any additional studies. Orwin's (1983) fail-safe N calculated 137 missing studies would be required to reduce Hedge's g to under 0.01.

Effect of Low-intensity CBT-based Interventions on Depression

Twelve studies evaluated the effect of a low-intensity CBT-based intervention on depression. Six studies used the Center for Epidemiological Studies Depression Scale (CES-D) (Gallagher-Thompson *et al.*, 2008a; 2008b; Au *et al.*, 2010; Dowling *et al.*, 2013; Kajiyama *et al.*, 2013; Blom *et al.* 2015), three used the Beck Depression Inventory (BDI) (Steffen, 2000a; 2000b; Beauchamp *et al.*, 2005), one used the BDI-II (Steffen and Grant, 2016), one used the Depression subscale of the Brief Symptom Inventory (BSI-D) (Chang, 1999), and one used the Geriatric Depression Scale (GDS) (Tremont *et al.*, 2008).

Data from Steffen (2000a) and Steffen (2000b) was analysed as a single composite study (Steffen, 2000), as described above, yielding a total of eleven studies used in the analysis. The individual study effects sizes as Hedge's g ranged from 0.17 (95% CI -0.24 to 0.59, $p=0.41$) to 0.74 (95% CI -0.05 to 1.54, $p=0.07$). In meta-analysis, there was a significant reduction of depression. The pooled effect size was 0.27 (95% CI 0.15 to 0.39, $p<0.01$) (Appendix K). The exclusion of Dowling *et al.* (2013) in sensitivity analysis did not alter the overall effect size ($g=0.27$, 95% CI 0.15 to 0.40, $p>0.01$) and hence it was not removed. There was no statistically significant heterogeneity found between studies ($I^2=0\%$ $p=0.98$).

Visual inspection of the funnel plot suggested possible publication bias (Appendix L). Duval and Tweedie's (2000) trim and fill method imputed one study estimating an adjusted Hedge's g of 0.26 (95% CI 0.14 to 0.38). Orwin's (1983) fail-safe N calculated 283 missing studies would be required to reduce Hedge's g to under 0.01.

Effect of Low-intensity CBT-based Interventions on Burden

Three studies evaluated the effect of a low-intensity CBT-based intervention on burden (Tremont *et al.*, 2008; Dowling *et al.*, 2013; Chiu *et al.*, 2015), using the Zarit Burden Interview (ZBI) (Zarit *et al.*, 1980). The individual study effects sizes as Hedge's g ranged from 0.32 (95% CI -0.21 to 0.85, $p=0.24$) to 0.84 (95% CI 0.15 to 1.54, $p=0.02$). In meta-analysis, there was a significant reduction of burden as measured by the ZBI. The pooled effect size was 0.53 (95% CI 0.16 to 0.90, $p=0.01$) (Appendix K). The exclusion of Dowling *et al.* (2013) in sensitivity analysis did not alter the overall effect size ($g=0.53$, 95% CI 0.02 to 1.03, $p=0.04$) and hence it was not removed. There was no statistically significant heterogeneity found between studies ($I^2=0\%$ $p=0.49$).

Visual inspection of the funnel plot suggested possible publication bias (Appendix L). Duval and Tweedie's (2000) trim and fill method imputed two studies estimating an adjusted Hedge's g of 0.32 (95% CI -0.04 to 0.67). Orwin's (1983) fail-safe N calculated 156 missing studies would be required to reduce Hedge's g to under 0.01.

Effect of Low-intensity CBT-based Interventions on Distress

Five studies evaluated the effect of a low-intensity CBT-based intervention on distress (Beauchamp *et al.*, 2005; Gallagher-Thompson *et al.*, 2008a; 2008b; Chiu *et al.*, 2015; Dowling *et al.*, 2013). The Caregiver Strain Instrument (CSI) (Bass *et al.*, 1998) was used by Beauchamp *et al.* (2005). All other studies used the Perceived Stress Scale (PSS) (Cohen and Williamson, 1988; Gallagher-Thompson *et al.*, 2008a; 2008b; Dowling *et al.*, 2013; Chiu *et al.*, 2015).

The individual study effects sizes as Hedge's g ranged from 0.23 (95% CI 0.00 to 0.46, $p=0.05$) to 1.01 (95% CI 0.18 to 1.83, $p=0.02$). In meta-analysis, there was a significant reduction of distress yielding a pooled effect size of 0.33 (95% CI 0.17 to 0.48, $p<0.01$) (Appendix K). A sensitivity analysis was conducted excluding Dowling *et al.* (2013), which resulted in a comparable pooled effect of 0.30 (95% CI 0.15 to 0.46,

$p < 0.01$), and hence it was not removed. There was no statistically significant heterogeneity found between studies ($I^2 = 0.00\%$ $p = 0.50$).

Visual inspection of the funnel plot suggested possible publication bias (Appendix L). Duval and Tweedie's (2000) trim and fill method imputed two studies estimating an adjusted Hedge's g of 0.29 (95% CI 0.12 to 0.46). Orwin's (1983) fail-safe N calculated 190 missing studies would be required to reduce Hedge's g to under 0.01.

Discussion

Caring for a person with dementia has a psychological impact on informal caregivers. The number of informal DCs is expected to rise and resources offering support are limited, particularly in less developed countries. CBT is an intervention which has shown some promise in reducing psychological difficulties in DCs, however CBT can be resource intensive. This study reviewed the effectiveness of low-intensity CBT-based interventions for informal DCs on the outcomes of anxiety, depression, burden and distress (stress/strain) compared to control conditions. Significant intervention effects were obtained for all psychological outcome variables, however the size of the effects varied across outcomes. Heterogeneity was low for all meta-analyses, though it is possible that the small number of studies did not allow for heterogeneity detection (Cuijpers, 2006). Possible publication bias was identified for depression, burden, and distress analyses, though adjusted effects sizes are not discussed as publication bias assessment is unreliable when less than 30 studies are included (Cuijpers, 2016).

Anxiety

Only five studies were included in the meta-analysis of anxiety outcomes, which is consistent with observations that anxiety is somewhat neglected in DC intervention literature (Schulz *et al.*, 2002; Li *et al.*, 2013). The overall effect of low-intensity CBT-based interventions on reducing anxiety was moderate with a Hedge's g of 0.58. This was reduced to a small effect of 0.35 following the exclusion of Villareal-Reyna *et al.* (2012) as

an outlier. Villareal-Reyna *et al.* (2012) involved the greatest number of facilitator contact hours and was the only study which employed a group-based intervention which reported on anxiety. It is possible that factors such as increased facilitator contact time, and contact with other DCs in the group setting may have resulted in a greater intervention effect, however further research would be required to explore these hypotheses.

Depression

An overall reduction in depression was found with a small effect size of 0.27 pooled across the twelve included studies. The obtained effect is smaller than reports of the effect of CBT for DCs when not restricted to low-intensity (Kwon *et al.*, 2017). It could be argued that low-intensity CBT-based interventions may only be beneficial for DCs with low levels of depression, and the studies included in this meta-analysis did not place limits on participant depression levels. However, in a non-caregiver population, participants with more severe depression do not benefit less from low-intensity interventions than those with milder depression (Bower *et al.*, 2013). It is therefore important for future research to examine which aspects of low-intensity CBT-based interventions have the greatest impact on depression outcomes.

Burden

Low-intensity CBT-based interventions were shown to reduce burden outcomes with a moderate effect size of 0.53. However only three studies were included in the analysis and as such results should be interpreted with caution. Although burden is not a clinical diagnosis, it is unexpected that so few studies examined burden, given reports of high levels of burden in DCs (Brodaty *et al.*, 2014; Liu *et al.*, 2017). It has been suggested that some burden predictor variables (e.g. person with dementia characteristics, care situation) are mediated by DC appraisals (Sörensen and Conwell, 2011). Given that low-intensity CBT-based interventions often target DC appraisals; it would be beneficial for future low-intensity CBT-based studies to include burden as an outcome.

Distress

An overall reduction in distress, defined as perceived stress or strain, was found across the five included studies with a small effect size of 0.33, though results should be interpreted with caution due to the small number of studies. The paucity of studies examining distress may be due to differing definitions of the variable (Ferrara *et al.*, 2008; Cress, 2009). It is therefore recommended that future low-intensity CBT-based DC intervention studies include measures of distress defined as perceived stress or strain to provide a more reliable estimate of effectiveness.

Strengths and limitations

A strength of this study is the rigorous selection strategy, and use of a clear definition for 'low-intensity'. However, the latter may also be considered a limitation, as there is no universal agreement regarding defining interventions as low-intensity. The examination of multiple outcomes is a strength, though this limited the depth of discussion for each outcome. Furthermore, it is important to note there are additional relevant DCs outcomes which were not included (e.g. life satisfaction) (Thomas *et al.*, 2006).

The small number of studies included in each meta-analysis created several limitations. Firstly, it may not have been possible to detect heterogeneity even if it existed (Cuijpers, 2016). As such, the impact of intervention characteristic moderators was not examined. Given the variety in intervention characteristics of included studies (e.g. intervention components, facilitators, delivery format etc.), it is not possible to conclude which low-intensity CBT-based interventions are most beneficial, how they should be delivered and by whom. Secondly, most studies recruited caregivers for people with Alzheimer's dementia, or did not specify dementia type. Therefore, it cannot be determined if low-intensity CBT is effective for caregivers of different dementia illnesses, as some types of dementia present with more behavioural problems which could lead to increased caregiver psychological difficulties (Riedijk *et al.*, 2006). Furthermore, all

included studies used samples from developed countries. This limits the generalisation of results to less developed countries. Caregivers in such areas are reported to have less knowledge of dementia, less formal support, greater financial difficulty and differing cultural needs (Kalula and Petros, 2011). It is not known if this combination of factors would alter the effectiveness of low-intensity CBT-based interventions.

Research Implications

This study highlights the requirement for larger scale low-intensity CBT-based intervention studies, using participant samples from both developed and less developed countries, reporting on a variety of outcomes relevant to DCs. The inclusion of long-term follow-up outcomes will help determine if effects for DCs are long lasting, or time limited, as has been suggested for low-intensity CBT-based interventions in other clinical populations (Ali *et al.*, 2017). Further research should aim to identify important characteristics of low-intensity CBT-based interventions which moderate outcomes, to aid the development of future interventions. It is also important that future studies report information about the profession and qualifications of intervention facilitators to support this process, as this review identified such details were frequently absent and required clarification from authors.

Clinical implications

Despite the above limitations, this study provides support for the use of low-intensity CBT-based interventions for DCs. The expected increase in the need for informal dementia care is likely to have a greater impact on DCs residing in areas where resources are limited. Therefore, a small or medium reduction in the psychological difficulties experienced by DCs is an important reduction. Particularly when a smaller amount resources are expended to achieve such an effect, as is the case with low-intensity CBT-based interventions. This study also provides support for further exploration into a stepped-care approach for DCs.

Conclusions

This study shows that low-intensity CBT-based interventions reduce levels of anxiety, depression, burden and distress (stress/strain) experienced by DCs, when compared to non-active controls. However, more research is required to increase the generalisability of findings to the poorly resourced populations which may benefit the most. Furthermore, additional exploration into the most important aspects of low-intensity CBT-based interventions is necessary to guide intervention development to increase effectiveness.

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Chapter 5. Thesis Portfolio Discussion

The first empirical paper included in this thesis portfolio aimed to quantitatively synthesize the literature on the prevalence of anxiety in caregivers for people with dementia. The second empirical paper aimed to establish the efficacy of low-intensity CBT-based interventions on the outcomes of anxiety, depression, burden and distress in dementia caregivers.

Dementia Caregiver Anxiety and the Efficacy of Low-intensity CBT-based Interventions

The first empirical paper identified a high prevalence of anxiety caseness in dementia caregivers. The pooled prevalence rate of 32.1 percent was greater than the reported 7.3 prevalence rate in the general population (Baxter et al., 2013). However relatively few studies ($n = 10$) were included in the analysis, which suggests anxiety is largely neglected in the dementia caregiver epidemiological literature, compared to depression (Collins & Kishita, 2018). Nonetheless, the obtained anxiety prevalence rate was comparable to the reported 31.2 percent prevalence rate for depression caseness in dementia caregivers (Collins & Kishita, 2018). This provided a strong rationale for including anxiety as an outcome measure in the second empirical paper.

In the second empirical paper, it was noted that only five low-intensity CBT-based interventions included anxiety as an outcome measure, which suggested anxiety is also neglected in the dementia caregiver intervention literature. The overall effect size for the efficacy of low-intensity CBT-based interventions on anxiety was moderate ($g = 0.58$), though this was reduced to a small effect ($g = 0.35$) when an outlier was excluded in sensitivity analysis (Villareal-Reyna et al., 2012). However, it is possible that Villareal-Reyna et al. (2012) was considered a statistical outlier due to the small number of studies included, and it may not have been considered an outlier if a greater number of studies were included. Whilst the publication bias assessment suggested there were no missing studies, it was noted the assessment was unreliable due to the small number of included

studies (Cuijpers, 2016). Regardless of whether the true effect size is small or moderate, it could be argued that the efficacy of low-intensity CBT-based interventions on anxiety in dementia caregivers is of clinical significance, given the high prevalence of anxiety in dementia caregivers, and the smaller amount of resources expended in delivering such interventions.

Low-intensity and high-intensity CBT-based interventions for anxiety

It was not possible to compare the pooled efficacy of low-intensity CBT-based interventions to the pooled efficacy of high-intensity CBT-based interventions for anxiety in dementia caregivers. This is because there is not currently a review of the evidence regarding the latter, which may be due to the relative neglect of anxiety as an outcome in dementia caregiver intervention studies.

However, a study by Losada et al. (2015) demonstrated a high-intensity CBT intervention reduced dementia caregiver anxiety symptoms, though the results were not statistically significant when compared to a control group. The same study demonstrated a high-intensity 'third-wave' CBT-based intervention, specifically, Acceptance and Commitment Therapy (ACT), achieved a statistically significant reduction in anxiety symptoms compared to the control group. It is therefore important that future high-intensity CBT-based interventions include anxiety as an outcome measure to gain a better understanding of high-intensity CBT-based interventions in their own right, but also to allow comparison with low-intensity CBT-based interventions. The ability to compare the efficacy of high and low-intensity interventions, and associated similarities and differences in intervention and participant characteristics may help to broaden our understanding of which CBT-based intervention characteristics are most effective for various subgroups of dementia caregivers. Understanding such factors may aid the development of a resource efficient stepped-care approach to supporting anxiety in dementia caregivers, so that the least intense but most effective intervention may be offered.

Moderators of anxiety prevalence in dementia caregivers

Whilst significant heterogeneity was found within the pooled estimate of anxiety prevalence in dementia caregivers, it was not possible to reliably examine factors which may moderate prevalence, due to the small number of included studies. Only one subgroup analysis was conducted, examining the moderating impact of measurement tool type (diagnostic vs. self-report symptom measure), though results were inconclusive.

Other potentially moderating factors could not be explored using subgroup analysis, due to the small number of included studies. There was too much variety within some potentially moderating factors, and therefore not enough studies within each subgroup to allow for comparison. For example, there was a wide variety of individual prevalence measurement tools (six different tools were used across ten studies), which meant there were few studies, and often only one study, in each subgroup. Conversely, some potentially relevant moderating characteristics contained too little variety within the study sample to permit subgroup analysis. For example, all but one included study were classified as using caregiver samples from very highly developed countries. Furthermore, the majority of studies used participants caring for a person with Alzheimer's dementia only, or used a mixed sample comprising a majority of Alzheimer's caregivers. This prevented examining the moderating effect of country development status and care recipient dementia type on the prevalence of anxiety in caregivers. Therefore, it was not possible to draw conclusions regarding which subgroups of caregivers may be more or less likely to experience anxiety, nor to make subsequent related clinical recommendations.

Moderators of the effect of low-intensity CBT-based interventions on anxiety in dementia caregivers

Moderator analysis was not conducted for the effect of low-intensity CBT-based interventions on anxiety in dementia caregivers, as statistically significant heterogeneity

was not present within the pooled effect estimate. However, it is important to note that it is not always possible to detect heterogeneity when the number of included studies is small. Indeed, the intervention efficacy analysis for anxiety included a sample size which was half the size of that which was included in the anxiety prevalence analysis.

It was however noted that a similar spread of study and participant characteristics was found across the studies included in the intervention analysis. Specifically, the studies all occurred in developed countries, and the most frequently included caregivers were those who cared for a person with Alzheimer's dementia, or the dementia type was mixed with a majority of Alzheimer's, or was not specified. Only one included study used a sample of caregivers for people with fronto-temporal dementia, though the study did not include anxiety as an outcome.

It is also worth noting that all included studies used CBT-based interventions which were defined as 'multicomponent', including both CBT-based and non-CBT-based components. However, this thesis portfolio is not able to comment on which components are most important. Furthermore, there were no 'third wave' type low-intensity CBT-based interventions included in the second empirical paper. Given the above mentioned findings of Losada et al. (2015), low-intensity ACT interventions for dementia caregivers may be worth future exploration.

In summary, it is important that both anxiety prevalence moderators, and moderators for interventions effectiveness on anxiety are identified. This will help identify caregivers which may be more vulnerable to experiencing anxiety, and allow interventions to be tailored by delivering the most effective components in the most effective manner.

Additional Outcomes Included in the Second Empirical Paper

Whilst anxiety was explored across both empirical papers, the additional outcomes of depression, burden and distress (stress/strain) were included in the second empirical paper, which examined the efficacy of low-intensity CBT-based interventions. These

outcomes had been selected and given equal importance to anxiety, based on the recommendations made by Schulz et al. (2002). To clarify, it was recommended that all dementia caregiver intervention studies include measures of clinical symptomology (e.g. depression; anxiety), quality of life (e.g. psychological wellbeing; life satisfaction), social validity (e.g. the acceptability of an intervention) and social significance (e.g. placement of care recipient in formal residential care) (Schulz et al., 2002). It was suggested that doing so would improve the ability to draw meaningful conclusions from the diverse range of dementia caregiver interventions and outcomes in the literature.

It was not unexpected that in the second empirical paper the most frequently examined outcome in the included studies was depression ($n = 12$), and that anxiety was less frequently included ($n = 5$), given the aforementioned neglect of anxiety in the literature. However, it was somewhat unexpected that so few studies examined burden ($n = 3$) given that burden is reported to be a prevalent difficulty (Collins & Kishita, 2018). The lack of studies examining distress as stress/strain ($n = 5$) could be described as somewhat more expected given a lack of literature synthesizing the prevalence of stress/strain in the dementia caregiver population. Nonetheless, the majority of included studies were conducted after Schulz's (2002) recommendations were made, and after prior intervention reviews have highlighted the importance of such variables (Pinquart & Sörensen, 2006). Therefore, it may have been expected that more studies would have included a wider range of the above outcome variables.

However, it should be noted that the definition of burden and stress or strain varies considerably within the caregiver literature. Some studies have used the phrase 'burden' as an umbrella term to describe perceived burden, and stress and strain, whereas other studies describe such variables as related but conceptually different concepts. The lack of clarity in the literature may contribute to the small number of low-intensity CBT-based intervention studies including burden and stress/strain as outcomes. Furthermore, it has been suggested that objective caregiver burden is less important than subjective burden

in terms of both caregiver and care-recipient outcomes (Zarit, Todd, & Zarit, 1986).

Therefore, it could be hypothesised that whether or not burden is included as an outcome may in part depend on the chosen definition of burden.

Social validity and social significance were not included as outcomes in the second empirical paper, however the outcomes were considered when rating the quality of included studies. Interestingly, only five out of 13 studies included a measure of social validity, usually presented as an 'acceptability of the intervention' survey (Beauchamp et al., 2005; Dowling et al., 2013; Kajiyama et al., 2013; Steffen, 2000; Tremont et al., 2008) (Appendix J). Only one study included a measure of social significance, reporting on the caregiver status of participants following the intervention (Steffen & Grant, 2016). It seemed that reporting on these outcomes was not associated with the total study quality rating, however a statistical analysis was not conducted. It is possible that whilst it would be beneficial for studies to include all of the outcomes as recommended by Schulz et al. (2002), doing so may increase the demand on participants and hence reduce the feasibility of the study.

Efficacy of Low-intensity CBT-based Interventions on the Additional Outcomes of Depression, Burden and Distress

In summary, low-intensity CBT-based interventions had a pooled small effect on reducing depression ($g = 0.27$) and distress (stress/strain) ($g = 0.33$), and a moderate effect on reducing burden ($g = 0.53$). It is important to remember that separate meta-analyses using different studies were conducted for each outcome. Therefore, caution should be exercised when comparing the efficacy of low-intensity CBT-based interventions on each of the outcome variables included in the second empirical paper.

Heterogeneity Across Studies

There was no statistically significant heterogeneity found in any of the meta-analyses in the second empirical paper, however as previously mentioned the small

number of studies included in each analysis may have prevented detection of heterogeneity if it existed (Cuijpers, 2016). Unfortunately, this meant subgroup analyses could not be performed to identify important moderator characteristics. As a result, it is difficult to draw conclusions regarding which participant and intervention variables are likely to impact on the above outcomes. This is important given the wide range of some characteristics of included studies (e.g. intervention component content), and the narrow range of others (e.g. care recipient dementia type, country development status).

It is particularly important to consider the wide range of intervention components used across different studies included in the meta-analyses. Whilst all studies were required to include components based on CBT, the majority of studies included additional non-CBT components which varied widely in content and delivery. However, as moderator analysis could not be conducted, we are not able to state which are the most important components. As such, we do not know if CBT components are those which produce the greatest effect, or which aspects of CBT are more effective for this population. Nor do we know the importance of the non-CBT components, or if particular combinations of CBT and non-CBT components are the most beneficial. It is also possible that common factors present across some interventions (e.g. meeting with a facilitator, meeting other caregivers in a group setting) may impact on the effect size. Whilst appropriately selected control groups may control for such factors within individual studies, it is important to consider the possible varying influence of such factors on the obtained pooled effects sizes.

Understanding how these intervention factors impact on dementia caregiver outcomes will help to tailor low-intensity CBT-based interventions. Redundant components could be eliminated to increase efficiency and allow resources to be focused on more effective components. This would likely benefit both caregivers and service providers.

Nonetheless, the second empirical paper provides initial support for the use of low-intensity CBT-based interventions for dementia caregivers, as even a small effect may be clinically significant for this population. Future research would benefit from exploring how low-intensity CBT-based interventions can be adapted to increase the size of the effects.

Low-Intensity CBT-based Interventions in the Context of a Theoretical Model of Dementia Caregiver Morbidity

The first empirical paper identifies anxiety as a prevalent difficulty in dementia caregivers, and the second empirical paper examines the impact of low-intensity CBT-based interventions on anxiety, depression, burden and distress. Whilst, neither paper is able to offer insight into the mechanisms underlying the difficulties experienced by dementia caregivers, it is possible to consider the results in the context of relevant models of dementia caregiver morbidity.

The Stress-Process model of caregiving was first proposed by Pearlin et al. (1990) and has been subsequently discussed and adapted for dementia caregivers (Hilgeman et al., 2009; Lavretsky, 2005; Richard Schulz & Martire, 2004). The model postulates that informal caregivers are exposed to primary stressors, such as caregiving tasks (e.g. personal care, housework, assisting the care recipient to attend medical appointments), care recipient behaviours which challenge (e.g. wandering, aggression, repetitive behaviour), and changes in the care recipient compared to prior to dementia, which may be experienced by the caregiver as a loss. Dementia caregivers are often subject to secondary stressors, as a direct result of primary stressors. For example, caregivers may experience a deterioration in relationships with other family members due to disagreements over caregiving commitments. Caregivers may experience challenges balancing caregiving demands with employment, and can experience financial difficulties. The combination of primary and secondary stressors is then appraised as demands in the context of perceived and actual resources and adaptive capabilities. The caregiver's perception of stress then mediates an emotional and behavioural response, or coping

style, which can then lead to or protect against psychological and physical morbidity (Garcia-Alberca et al., 2013; Snyder et al., 2015).

Furthermore, the caregiver's response to the perceived stress can subsequently influence primary and secondary stressors, either by inadvertently reinforcing difficulties, or by reducing them. For example, a caregiver who perceives themselves to be unable to cope with primary and secondary caregiving stressors, may respond to a care recipient's challenging behaviour with high levels of negative expressed emotion, which may in-turn increase care recipient distress and challenging behaviour (Li & Lewis, 2013). An overview of the model is presented in Figure 4.

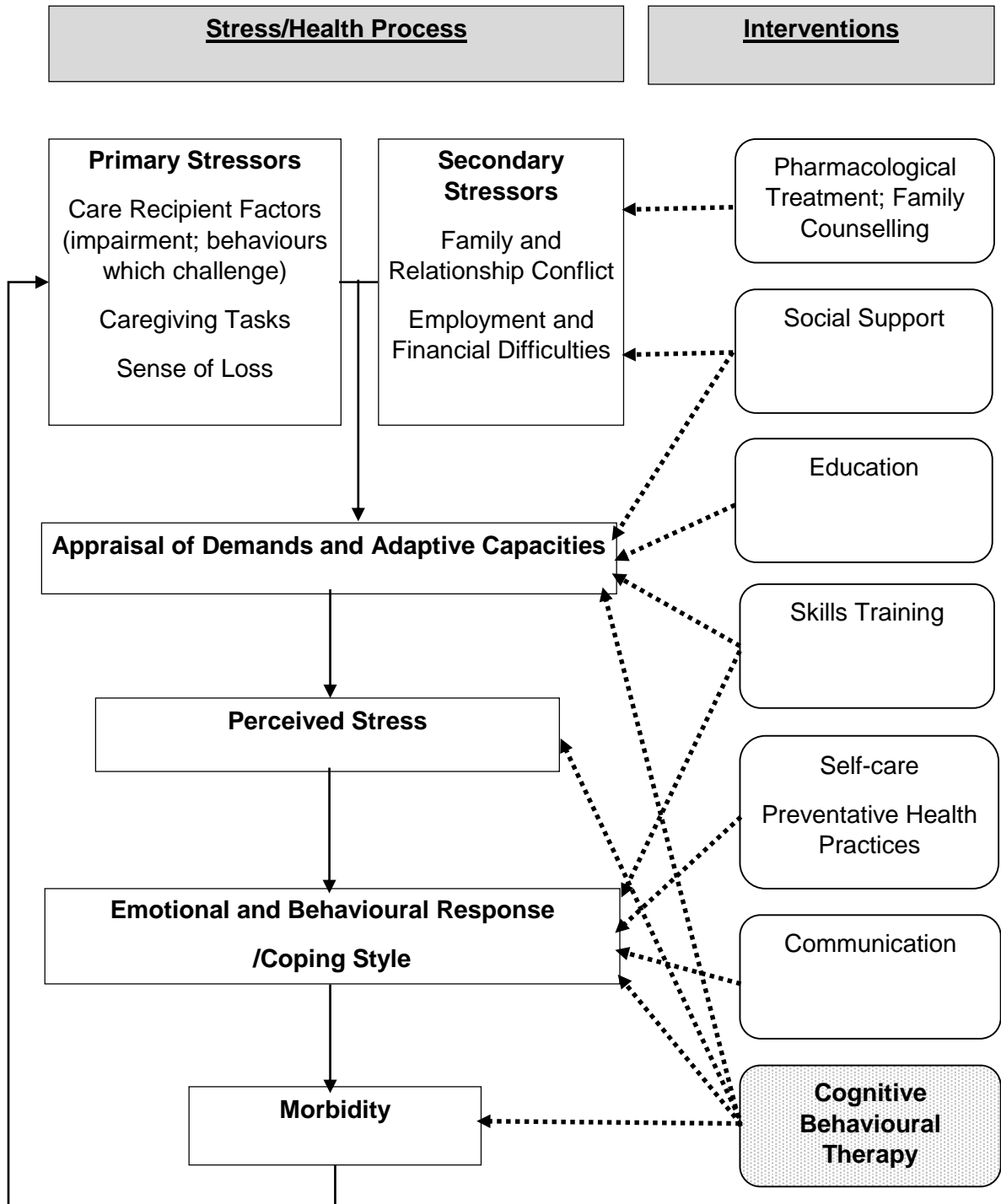


Figure 4. Stress-Process Model adapted from Schulz and Martire (2004)

Note. Does not include all dementia caregiver interventions.

It follows that interventions which target any aspect of the model are likely to have an impact on caregiver morbidity. This may explain the wide range of interventions currently in existence for reducing dementia caregiver morbidity (Pinquart & Sörensen, 2006a). For example, respite care may reduce primary stressors and increasing social support may alleviate secondary stressors. Education about communication and caregiver skills training may increase a caregiver's adaptive capacity, and alter behavioural responses to perceived stress. CBT is likely to impact several parts of the model, by altering caregiver appraisals and perceptions of stress, promoting the development of adaptive coping strategies or behavioural/emotional responses, and targeting specific psychological morbidity difficulties (e.g. depressive/anxious rumination) (Laidlaw, 2015). As discussed in the second empirical paper, low-intensity CBT aims to deliver CBT using less resources, and resource conservation is important when considering dementia caregiver interventions.

It was noted that all but two of the low-intensity CBT-based interventions included in the second empirical paper were classified as 'multi-component', containing both CBT-based and non-CBT based components. This is congruent with reports that multi-component interventions are the most effective (Pinquart & Sörensen, 2006a). It can be hypothesised that the effect of multi-component intervention is due to their action on several parts of the Stress-Process model. However, the small number of purely CBT-based low-intensity intervention studies, and lack of any non-active CBT psychoeducation low-intensity intervention studies included in the analysis, means it is not possible to draw conclusions about the relative importance of CBT components. Nor was it possible to identify which 'non-CBT-based' components were most important, due to the wide variety of components included across studies. Future research would benefit from exploring this further.

Summary of Thesis Portfolio Research Recommendations

This portfolio highlights several gaps in the current dementia caregiver literature. As such, recommendations for future research are discussed below, based on both of the empirical papers contained within this portfolio.

Anxiety as an overlooked psychological difficulty

Firstly, anxiety is a somewhat neglected psychological outcome in the literature. It would be beneficial for large scale anxiety prevalence studies to be conducted. It is important that heterogeneity is explored with a focus on identifying relevant care recipient, caregiver, and situational/environmental moderating factors. This will help to identify which caregivers may be more vulnerable to anxiety, and which may be more protected. This may allow for earlier intervention, and the distribution of limited resources to where they are needed the most.

Furthermore, given the relative high prevalence of anxiety caseness in dementia caregivers, which was found to be comparable to the reported prevalence of depression caseness in dementia caregivers, anxiety should be afforded parity with depression as an outcome of interest in intervention studies (Collins & Kishita, 2018).

Bridging the gap between prevalent psychological difficulties and effective interventions for dementia caregivers

It is important that future research explore factors which mediate prevalent psychological difficulties in dementia caregivers. Whilst there is literature exploring mediators for depression and burden, anxiety and perceived stress are less frequently examined and overall there is little clarity. Achieving a better understanding of factors which mediate such difficulties will help provide a clear rationale and clear targets for intervention development. Furthermore, understanding the relationships between the various psychological difficulties experienced by dementia caregivers will be beneficial, as for example, it is not currently clear why a majority of dementia caregivers who experience

depression, also experience comorbid anxiety, yet the reverse relationship does not exist (Mahoney, 2005). Given that comorbid depression and anxiety is associated with greater morbidity and poorer outcomes, it is important that this is investigated further.

It is important to note that the Stress-Process model described above does not differentiate between the different psychological difficulties associated with dementia caregiving, and instead refers only to 'morbidity'. It is expected that gaining a better understanding of mediating factors will allow for models of dementia caregiver difficulties to include a greater focus on psychological outcomes. It is also important that both factors relating to specific psychological difficulties (e.g. anxiety; depression) and common transdiagnostic processes (e.g. rumination, cognitive bias) are considered to aid the refinement and effectiveness of interventions. Understanding transdiagnostic processes may be particularly beneficial, given the often limited support resources available to dementia caregivers. For example, it may not always be possible for 'disorder' or 'problem' specific interventions to be offered, and focusing on relevant transdiagnostic processes may allow a greater number of caregivers to benefit from psychological interventions when delivered at a lower-intensity. However further research is required to test this hypothesis.

Understanding moderators in low-intensity CBT-based interventions

Caregiver and intervention characteristics moderators

As previously mentioned, this thesis portfolio was unable to identify factors which moderate the efficacy of low-intensity CBT-based interventions. Future research should aim to answer this question, with a focus on both caregiver (e.g. dementia type; hours caregiving; country etc.) and intervention variables (type of low-intensity CBT-based intervention; duration; facilitator qualification and contact time etc.), and should give consideration to a possible interaction between moderators. For example, it is not known how low-intensity CBT-based interventions are best delivered, or if they should be

delivered differently for different subgroups of caregivers, to achieve the best outcomes. As such, a greater number of large scale low-intensity CBT-based intervention RCTs would be beneficial.

Types of low-intensity CBT-based interventions

The systematic search in the second empirical paper identified mostly 'multi-component' low-intensity CBT-based interventions. It is not clear how the efficacy of 'multi-component' interventions compares to interventions using only CBT-based components, or indeed to low-intensity 'third-wave' CBT interventions such as ACT. Future research may wish to address this question. Furthermore, the second empirical paper did not identify any low-intensity CBT-based interventions of any type as being trialled in the United Kingdom (UK), or in less developed countries. Therefore, it is also recommended that low-intensity CBT-based intervention studies are replicated both in the UK, and in less developed countries.

Tailoring multi-component low-intensity CBT-based interventions

A wide range of additional components were used in the multi-component interventions included in the second empirical paper. However, it is not clear which non-CBT-based components are best included alongside the CBT-based components. Establishing the most important components will allow interventions to achieve better outcomes using less resources, by only delivering the most effective components.

It is possible that certain components may be important for all dementia caregivers, and some may be more effective for particular subgroups of caregivers (e.g. based on dementia type, ethnicity, etc. and other caregiver variables). However, given that dementia caregivers can be considered a highly heterogeneous group, it is possible that the most effective components are individual to each caregiver's unique characteristics and situation. Indeed, it has previously been recommended that dementia caregiver interventions offer individually tailored support (Pinquart & Sörensen, 2006). If

this is the case, it is important that a resource efficient method for assessing a caregiver's individual needs is developed, as given resource limitations, it is unlikely that each caregiver will be afforded an individual psychological assessment and formulation. Therefore, future research should also consider developing an effective and efficient method of assessing individual dementia caregiver needs, so that interventions may be adapted accordingly in an efficient manner.

Integrating knowledge of the positive aspects of caregiving into intervention research

The current state of the dementia caregiver literature, and indeed this thesis portfolio, tends to focus on the negative aspects of caregiving. For example, exploring caregiver psychological and physical morbidity, as well as examining potentially negative outcomes for the cared-for person (e.g. placement away from home in residential care) and society (e.g. financial burden of funding residential care) (Brodaty & Donkin, 2009; Schulz & Sherwood, 2008; Sörensen & Conwell, 2011). This may reflect a dominance of the application of a 'medical' or 'disorder' model to understand and potentially pathologise the experiences of dementia caregivers as difficulties requiring 'treatment' (Wampold & Imel, 2015).

It follows that the focus of interventions is frequently on reducing the negative aspects of caregiving and associated risk factors (Griffin et al., 2015; Pinquart & Sörensen, 2006). Whilst this often incorporates increasing protective factors, or building resilience against the negative aspects of caregiving, the potential positive aspects of the caregiving experience are often somewhat neglected (Dias et al., 2015; Liu et al., 2017).

One could argue that it is nonetheless important to consider the negative aspects of caregiving due to the reported impact on relevant stakeholders (e.g. caregiver, cared-for person, health-care providers, wider society etc.) (Alzheimer's Society, 2014). However, developing an understanding of and building on the positive aspects of

caregiving may be an additional strategy to achieve the same goal: to support the dementia caregiver population and relevant stakeholders. Focusing only on reducing the negative aspects could be considered a missed opportunity and neglectful of caregivers who would not describe their experiences of caregiving as distressing or burdensome. For example, a recent review identified four key positive aspects of caregiving for a person with dementia: a sense of accomplishment, an improvement in the cared-for person and caregiver relationship, an increase in family cohesion, and a sense of personal growth and purpose (Yu, Cheng, & Wang, 2018). It was identified that caregivers who experience a greater amount of positive aspects of caregiving are less likely to experience psychological/physical morbidity, and the cared-for person is less likely to require residential care (Yu et al., 2018).

Therefore, developing a better understanding of, and incorporating knowledge regarding why some caregivers don't develop difficulties (e.g. anxiety, depression etc.), and even thrive, is an important area for future research. As such, it is recommended that future dementia caregiver intervention research consider both reducing the negative aspects and supporting the positive aspects of caregiving. It is hypothesised that doing so may maximise positive outcomes.

Furthermore, greater consideration of the positive aspects of caregiving in interventions may aid the adaptation of interventions for cultural and ethnical differences in dementia caregiver experiences (Napoles, Chadiha, Eversley, & Moreno-John, 2010). For example, it has been suggested that some non-Western cultures experience greater positive aspects of caregiving than Western dementia caregivers (Roth 2015). It is possible that adopting an approach focused more on nurturing the positive aspects of caregiving may be more appropriate than focusing on reducing negative aspects, for caregivers who experience more positive aspects of caregiving.

Summary of Thesis Portfolio Clinical Implications

This thesis portfolio has several clinical implications. Whilst some clinical implications have been discussed above, an overview of the key implications are discussed here, along with additional clinical recommendations.

Firstly, it is important to note that the UK national clinical guidance regarding dementia is being updated. The current draft is due to be published in June 2018 and it recommends that clinicians are aware that informal carers are at an increased risk of depression, but the guidance neglects to mention caregiver anxiety (The National Institute for Health and Care Excellence, 2018). However, this thesis portfolio identified that informal dementia caregivers are much more likely to experience clinically relevant anxiety compared the general population, and at a prevalence rate similar to depression in dementia caregivers (Baxter et al., 2013; Collins & Kishita, 2018). Therefore, it is recommended that clinicians working with people with dementia also offer assessment and support for caregiver anxiety, and do not focus only on depression when considering psychological morbidity.

Furthermore, the high prevalence of dementia caregiver psychological morbidity highlights the importance of ensuring services offer psychological interventions for caregivers. Informal caregivers reportedly provide an equivalent of 11.6 billion pounds worth of care per year in the UK, and correspondingly high amounts are recorded worldwide (Alzheimer's Society, 2014; Jakobsen, Poulsen, Reiche, Nissen, & Gundgaard, 2011). Therefore, it is important that caregivers are supported both for their own wellbeing, but also due to the wider social and economic impact. This is imperative given the expectation that the prevalence of dementia, and as such need for informal care, is expected to rise (Prince et al., 2013).

This portfolio provides initial support for the use of low-intensity CBT-based interventions in reducing anxiety, depression, stress/strain and burden in dementia caregivers. However, it was noted that the national guidance in development does not

comment on the intensity of caregiver interventions, which professionals should facilitate interventions, and how long they should last (The National Institute for Health and Care Excellence, 2018). Given that services may not have sufficient resources, or may not be commissioned to offer high-intensity interventions, it is suggested that intervention intensity is considered in future guidance updates, so that clinical services may make informed decisions. It is also recommended that clinical services worldwide do not allow resource constraints to act as a complete barrier to offering psychological interventions for dementia caregivers, given the emerging support provided here for low-intensity CBT-based interventions.

Finally, it is recommended that clinical services already providing low-intensity CBT-based interventions, or indeed other interventions for dementia caregivers as part of standard care, consider recording outcome data and conducting and publishing service evaluations. Whilst not as highly regarded as RCTs, service evaluations can add to the evidence base and aid the process of achieving clarity, in the absence of a greater number of RCTs.

Critical Appraisal of the Thesis Portfolio

This thesis portfolio presented two original research papers focusing on people who informally care for a person with dementia. Understanding and supporting the difficulties associated with dementia caregiving is an area of research of great national and global importance. It can be considered a strength of this thesis portfolio that it has contributed to the evidence-base in this area. Furthermore, this portfolio has direct clinical relevance, and it is expected that publication of both empirical papers will allow clinicians to utilise the findings. However, it was noted that this portfolio elicits several unanswered questions, regarding both empirical papers. Whilst this may be considered a limitation of the portfolio, it is important to acknowledge that highlighting important gaps in the literature is often necessary to justify the direction of future research.

Critical appraisal of methodology

Both of the empirical papers contained within this portfolio adopted a meta-analysis methodology to quantitatively synthesize existing research. This method was arguably the most appropriate method to answer the research questions. However, it is important to acknowledge the potential value in adopting a qualitative approach to provide a richer understanding of dementia caregivers' experiences of both anxiety and low-intensity CBT-based interventions.

In general, the process of conducting a meta-analysis requires that several decisions are made, and each decision can introduce a degree of bias (Borenstein et al., 2009). With reference to this portfolio, such decisions include but are not limited to: the psychological outcomes selected and corresponding definitions used; the definition of low-intensity CBT-based interventions, and intervention subgroups; and the selection and adaptation of quality instruments.

Psychological outcomes

In the first empirical paper, anxiety was defined as caseness level of symptoms, or the presence of a psychiatric diagnosis. The decision was made to combine studies using a variety of measurement tools, and the appropriate statistical measures were taken to account for this by calculating the effect size as Hedges *g*. Nonetheless, had a single measurement tool been selected, it may have been possible to obtain a more reliable pooled prevalence estimate using a fixed-effects model as opposed to random-effects meta-analysis (Borenstein et al., 2010). However, it is likely that the number of included studies would be incredibly small, and a random-effects model would still be conducted due to the expected variation across study participant samples.

In the second empirical paper, multiple measurement tools were combined for each outcome, and several outcome variables were included. Anxiety and depression were used as measures of clinical symptomology, and burden and distress (defined as stress/strain) were used as measures of psychological wellbeing. However, the

terminology used to describe these variables contains some inconsistencies in the wider caregiver literature. Unsurprisingly, the inconsistencies appear most prevalent for burden and distress, as these terms do not directly relate to diagnosable disorders, as is the case with anxiety and depression, which are arguably well-defined phenomena.

The second empirical paper used the term 'distress' as a label for perceived stress or strain, which is described as the degree to which life events (stressors, or objective stress) are perceived as stressful (Cohen et al., 1983; Nübling, Stöbel, Hasselhorn, Michaelis, & Hofmann, 2006). However, 'distress' is often used as an umbrella term to describe a wider variety of psychological variables, including clinical symptomology and psychological wellbeing variables (Brodaty, Green, & Koschera, 2003; Ervin et al., 2015; Mourik et al., 2004; Pellegrino et al., 2010). Therefore, despite 'distress' being defined as perceived stress or strain in the methodology of the second empirical paper, it is possible that the label will be misinterpreted based on a reader's prior understanding of the term.

Furthermore, in the second empirical paper 'burden' was defined based on Zarit *et al.*'s (1980) definition. This describes burden as a caregiver's experience of the impact of caregiving demands on a carers health, wellbeing, finances, social life and relationship with the care-recipient (Zarit et al., 1980). It has been suggested that Zarit *et al.*'s definition of caregiver burden encompasses the constructs of both caregiver role strain, and personal strain, with a greater emphasis on the former (Hébert, Bravo, & Prévile, 2000). This would suggest that the concept of 'burden' overlaps with perceived stress/strain, or personal strain as it labelled above, but includes the additional construct of caregiver role strain.

However, some studies appear to use the terms 'burden' and 'perceived stress' as broad terms which are interchangeable (Gallagher-Thompson et al., 2008; Jones, Edwards, & Hounsome, 2012). Elsewhere, the definition of burden is further refined into 'psychological burden', 'physical burden', 'social burden' and 'emotional burden' (Ferrara et al., 2008). These terms respectively refer to a sense of a loss of opportunity, negative

feelings about one's own health, a sense of negative familial and colleague relationships, and negative feelings towards to the care-recipient (Ferrara et al., 2008).

Overall, burden as a construct lacks conceptual clarity across the caregiver literature. Therefore, despite having defined burden and distress in the second empirical paper, the use of arguably ambiguous outcomes may lead to a misinterpretation of the results when considered in the context of the variety of definitions used within the wider literature. As such, the generalisability of the findings should be conducted with consideration to the specific definitions detailed in the second empirical paper. Furthermore, comparing the results of the second empirical paper to other studies focusing on outcomes labelled as burden, distress, stress or strain, will need to be executed with caution, given the possibility for differing definitions.

Definition of low-intensity CBT-based interventions and intervention subgroups

The definition of low-intensity CBT-based interventions was based on a public health stepped-care model for anxiety and depression, and existing review literature on low-intensity interventions (The National Institute for Health and Care Excellence, 2009, 2011a, 2011b; Richards et al., 2010; Rodgers *et al.*, 2012). Nonetheless, the definition could be considered somewhat arbitrary as there is no universally accepted cut-off for describing interventions as low or high-intensity. The intensity of an intervention could even be considered relative to the particular mix of professionals and resources available to individual clinical services.

With regards to the low-intensity CBT-based intervention subgroups described within the second empirical paper, it is important to note the somewhat broad definition of what constitutes 'third-wave' CBT-based interventions. Had any 'third-wave' CBT-based intervention studies been included in the analysis, it may have been necessary to further explore the chosen definition in the discussion. It is also important to highlight the selected

definition for 'CBT-based psychoeducation' interventions were described as any passive intervention providing CBT-based education. However, the term 'psychoeducation' is often used to describe interventions which provide information about dementia, and how caregivers can respond to the various dementia-related difficulties (The National Institute for Health and Care Excellence, 2018). Whilst 'CBT-based psychoeducation' was clearly defined within the methods section of the second empirical paper, the results may be misleading if the distinction is not noted by the reader.

Selection and adaptation of quality instruments

Differing measures of study quality were used for the first and second empirical paper, reflecting the differing methodologies. Both instruments were adapted to reflect important quality characteristics specific to each empirical paper, and some items were more clearly defined to increase the objectivity of scoring. However, whilst the adapted instruments were piloted by the researchers involved in this portfolio, the reliability and validity of the adapted instruments was not robustly examined. Furthermore, the adaptations limit the ability to compare the quality of the included studies, to studies which are included in other reviews and meta-analyses and have been rated using the original instruments.

Furthermore, it is worth noting that for pragmatic reasons, the quality of included studies in the first empirical paper was independently rated and subsequently agreed between researchers for only ten percent of the studies. It could be argued that Cohen's Kappa coefficient should have been calculated to provide an estimate of inter-rater reliability, however, Cochrane guidelines advise against this due to arbitrary guidelines regarding the numerical values and associated descriptive levels of inter-rater reliability (Higgins & Green, 2011). Ideally, two independent raters would have completed the quality assessment for all studies, as was the case in the second empirical paper.

Critical appraisal of results

The number of eligible studies included in both empirical papers can be described as relatively low, and a-priori power calculations were not conducted. This created several limitations. Firstly, the overall power of the analyses to detect significant effects was likely reduced as a result of a small sample. This results in a greater likelihood of not detecting a significant effect when it fact it may exist, which is described as making a Type-II error (Banerjee, Chitnis, Jadhav, Bhawalkar, & Chaudhury, 2009). The impact of a low sample size on power tends to be greater in random-effects meta-analyses compared to fixed-effects analyses, and the former was employed in both empirical papers (Jackson & Turner, 2017).

Secondly, the impact of a small sample size was greater during subgroup analyses, as each subgroup contains a sample of studies which is smaller than the total sample. Indeed, the subgroup analysis conducted in the first empirical paper failed to find a significant pooled prevalence estimate for one sub-group, which could be the result of a Type-II error. Thirdly, in both empirical papers the small sample size did not allow for heterogeneity to be adequately explored through all of the planned subgroup comparisons. As such, heterogeneity could not be explained which led to several unanswered questions regarding moderator variables, and limited the reliability and generalisability of the results. Furthermore, the small number of studies in both empirical papers reduced the reliability of the publication bias assessment, and as a result it was not possible to reliably estimate if such bias existed.

The eligibility criteria for each empirical paper could have been broadened to achieve a greater number of included studies. However, this likely would have introduced a greater amount of methodological bias by combining potentially radically different studies. This type of bias is often described using the analogy of attempting to combine 'apples' and 'oranges' (Higgins & Green, 2011).

The decision to proceed with a quantitative synthesis when faced with a small number of eligible studies could be challenged. However, there are no definitive

guidelines regarding what is considered the minimum number of studies needed to quantitatively combine effects in meta-analysis, and the recommended minimum number of studies is as low as five (Cuijpers, 2016; Higgins & Green, 2011; Jackson & Turner, 2017). Additionally, continuing with a quantitative synthesis was deemed clinically relevant for both empirical papers. It should also be noted that the first empirical paper contained a greater number of studies than an earlier synthesis of anxiety prevalence, and the number of studies included in the second empirical paper was comparable to meta-analyses of other interventions for dementia caregivers (Boots et al., 2014; Kwon et al., 2017).

Overall Thesis Portfolio Conclusions

This thesis portfolio demonstrates anxiety is a prevalent psychological difficulty experienced by dementia caregivers. As such, anxiety should be afforded as much focus as depression in the dementia caregiver literature, both in terms of developing an understanding of moderating factors and underlying mediating mechanisms, and as an outcome of interest in interventions studies. Furthermore, there is initial support for low-intensity CBT-based interventions for reducing anxiety, depression, burden, and stress/strain in dementia caregivers. Further research is required to establish which aspects of low-intensity CBT-based interventions are most effective and how they are best delivered, to maximise efficacy whilst minimising the expenditure of resources.

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Appendix A

International Psychogeriatrics Journal Submission Guidelines

Instructions for contributors

International Psychogeriatrics

Reviews of the Literature: Authors intending to submit a literature review should check recent issues of *International Psychogeriatrics* to ensure that no review of the topic they propose to discuss has been published in the journal in recent times. Review articles may have up to 75 relevant references. Authors contemplating the submission of a literature review article are welcome to contact the editor to discuss the appropriateness of the topic prior to submission (ipaj-ed@cambridge.org). Literature reviews should have an abstract.

Organization and style of manuscripts

Title page and corresponding author: Each article must have a title page with the title of the article, a list of all authors and their titles, affiliations and addresses. Each author must select only ONE country as their location. Author qualifications should not be listed as these are not published in the journal. The title page should explicitly identify the author to whom correspondence about the study should be addressed and that author's email address, telephone number, fax number and postal address must be clearly stated.

Abstract (Structured): Abstracts for original research and reviews should be structured and incorporate the following headings: Objectives, Design, Setting, Participants, Intervention (if any), Measurements, Results, and Conclusions. Abstracts should communicate the primary findings and significance of the research. They should not exceed 250 words in length. Abstracts for brief reports should not exceed 200 words.

Keywords: Under this heading and beneath the abstract, please list up to 8 words for the purpose of indexing.

Running title: This should contain no more than 50 characters including spaces.

Introduction: Briefly state the relevant background to the study to provide the necessary information and context to enable non-specialists to appreciate the objectives and significance of the paper. Most introductions to articles received for review are too long.

Methods: Materials and procedures should be described in sufficient detail to enable replication. Any statistical procedures used should be outlined and their use should be justified here. Results should not be included in the Method(s) section. If statistical procedures are used, they should be described here in adequate detail. Choice of statistical technique should be justified including some indication of the appropriateness of the data for the technique chosen. Adequacy of the sample size for the statistical technique(s) used must be addressed. If appropriate, a description of the statistical power of the study should be provided. If multiple univariate significant tests are used, probability values (p-values) should be adjusted for multiple comparisons, or alternatively a multivariate test should be considered. Significance results (p values) must be presented with accompanying statistics.

Results: This section may contain subheadings. Authors should avoid mixing discussion with the results. Sample sizes should be delineated clearly for all analyses. Some indicator of variability or sampling error should be incorporated into the reporting of statistical results (e.g. standard deviation). Wherever possible an indicator of effect size (e.g. Cohens d, η^2 , Cramers V, 95% confidence interval) should be reported in addition to p values. If multiple univariate statistical tests are used p values should be adjusted for multiple comparisons or alternatively a multivariate test should be used. Obtained statistical values for tests should be reported with degrees of freedom (e.g. t, F, χ^2). Terms such as prevalence, population, or control group, should be used appropriately in the scientific sense.

Discussion: Interpretation of the results with respect to the hypothesis(es) and their significance to the field should be discussed here. Results should be interpreted in the light of the size of the effect found and the power

of the study to detect differences. Any methodological and other weaknesses of the study should be outlined, including limitations imposed by sample size. Careful consideration of the conclusion(s) for accuracy and alternative interpretation, and possible conflicts or resolution of conflicts in the field is encouraged. Limited speculation and directions for future research can be included.

References: For review papers, no more than 75 articles that have been published or are in press should be cited; for regular research articles no more than 40 references, for brief reports no more than 15 references, for commentaries and editorials no more than 10 references, and for letters no more than 5 references. Unpublished data, personal communications, and manuscripts submitted for publication should be cited in the text and the supporting material submitted with the manuscript. *International Psychogeriatrics* uses the **Harvard referencing system**. Within the text of each paper journal articles should be cited in the style (Smith and Jones, 1999). Where an article quoted in the body of the text has more than two authors the term "et al." should be employed, i.e., (Smith et al., 1999). Text citations of multiple articles should be separated by semicolons, i.e., (Smith and Jones, 1999; Smith et al., 1999). At the end of each paper, all cited references should be listed alphabetically in the style indicated below. If the Digital Object Identifier (doi) is known, it should be added to the reference.

Reference examples:

For a journal article: **Smith, J., Jones, W. I. and Doe, J. T. (1996).** *Psychogeriatrics for pleasure and profit: an expanding field.* *International Journal of Unreproducible Results*, 3, 240-242.
doi:12.3456/5123456789.

For a book: **Smith, J.A., Brown, P.Q., Jones, H.A. and Robinson, D.V. (2001).** *Acute Confusional States.* New York: Cambridge University Press. For a book chapter: **Park, K., Tiger, B. and Runn, F. (1999).** *Psychogeriatrics in context.* In G. Verdi and A. Boito, (Eds.) *New Medical Specialties* (pp. 240-260). Cambridge: Cambridge University Press.

Where an article or book chapter has more than six authors only the first author's name should be given followed by the words "*et al.*".

For further examples of reference style see papers in recent issues of *International Psychogeriatrics*.

Figures/Tables: The manuscript should contain **no more than five figures or tables** (no more than two figures or tables for brief reports). The copies submitted with the manuscript must be of sufficient quality to enable reviewers to evaluate the data. The journal has a small budget to permit some color to be printed in some issues but authors wishing to publish figures requiring color to communicate the data may be required to pay some or all the additional cost.

Figure/Table legends: Each caption should begin with a brief description of the conclusion or observation provided in the figure. These should be submitted as a separate section after the References.

Word limits: The text of Review articles should not exceed **5,000** words, Regular research articles 3,500 words, brief reports 1,500 words, commentaries and editorials 1,000 words, and letters 500 words. The text excludes title page, abstract, acknowledgements, references, tables, and figures. Articles may contain supplementary material which is published online only.

Comprehensive instructions for contributors are available at:
<https://www.cambridge.org/core/journals/international-psychogeriatrics>

Appendix B

Additional Item Definitions for Prevalence Critical Appraisal Instrument (PCAI)

The PCAI was used to critically appraise the quality of included studies. The use of the PCAI was conducted using the comprehensive guidelines included in Munn et al.'s (2014) paper. However, some items were further defined for the purposes of this study, as described below.

Item 1. Was the sample representative of the target population?

This study aimed to estimate the prevalence of anxiety in dementia caregivers, without placing restrictions on caregiver demographic details or characteristics, as described in the study method. Therefore, for the purpose of this study, this item was rated 'yes' if the study sought a sample with a demographic/characteristic mix congruent with literature examining characteristics of dementia caregivers (Alzheimer's Society, 2015; Brodaty & Donkin, 2009; Richard Schulz & Martire, 2004).

For example, a study sample would include: (a) A mix of adult and older adult caregivers (>65years old), with a majority of older adult caregivers. (b) A majority of, but not exclusively, female caregivers. (c) The cared-for person dementia type should include a majority of people with Alzheimer's disease, but not exclusively, and should include other types of dementia.

The item was rated 'no', if a study sample placed restrictions on a particular caregiver/cared-for person characteristic, to obtain a 'sub-sample' of the dementia caregiver population. For example, only caregivers for people with severe dementia, or only caregivers dwelling in inner-city areas.

Item 4. Were the study subjects and setting described in detail?

There is a lack of evidence regarding which participant and setting variables are associated with anxiety caseness (Cooper *et al.*, 2007). As such, it is not known which

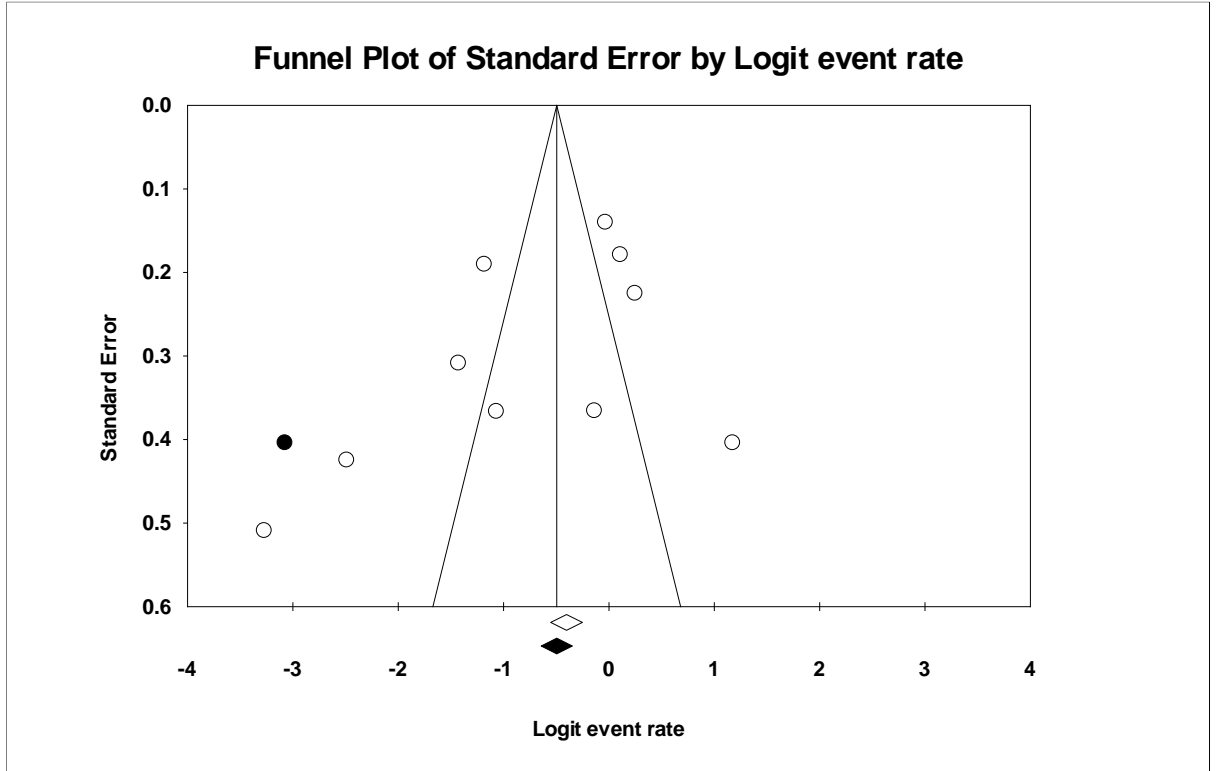
characteristics are more critically included in anxiety prevalence studies. Therefore, for the purposes of this study, this item was rated as 'yes' if a study described over half of twelve characteristics reported to be potentially important in dementia caregiver outcomes (age; gender; relationship to caregiver; economic status; ethnicity; education; subjective physical health; use of medication; living arrangement; care-recipient level of impairment; hours spent caregiving; months acting as a caregiver), with at least one item to be the number of hours caregiving or the number of months acting as a caregiver (Covinsky *et al.*, 2003; Cooper *et al.*, 2007; Cosseddu *et al.*, 2013; Liu *et al.*, 2017).

Item 9: Confounding factors/subgroups/differences identified and accounted for?

There is a lack of conclusive evidence regarding confounding factors and moderators for anxiety prevalence in dementia caregivers (Cooper, Balamurali, & Livingston, 2007). Therefore, for the purposes of this study, this item was rated as 'yes' if over half of the characteristics mentioned above as relevant for item 4 (age; gender; relationship to caregiver; economic status; ethnicity; education; subjective physical health; use of medication; living arrangement; care-recipient level of impairment; hours spent caregiving; months acting as a caregiver) were accounted for in the analysis.

Appendix D

Publication Bias Assessment Funnel Plot for Anxiety Prevalence Studies



Appendix E

Complete Search Terminology Used in the Systematic Literature Search and Adaptations for Each Source

Full Search Terminology

(a) Dement* OR Alzheimer* OR "Lewy bod*" OR "fronto*temporal" OR "LBD" OR "DLB" OR "FTD" OR "progressive cognitive impairment" OR "progressive cognitive decline" OR "progressive cognitive illness" OR neurodegenerative

AND

(b) Carer* OR caregiver* OR care OR caring OR famil* OR spouse* OR partner* OR relative*

AND

(c) "Cognitive behavio*ral therapy" OR CBT OR "cognitive therapy" OR "acceptance and commitment therapy" OR "ACT" OR "metacognitive therapy" OR "mindfulness*based cognitive therapy" OR "MBCT" OR "behavio*ral activation" OR "behavio*ral therapy" OR "exposure therapy" OR "systematic desensiti*ation"

AND

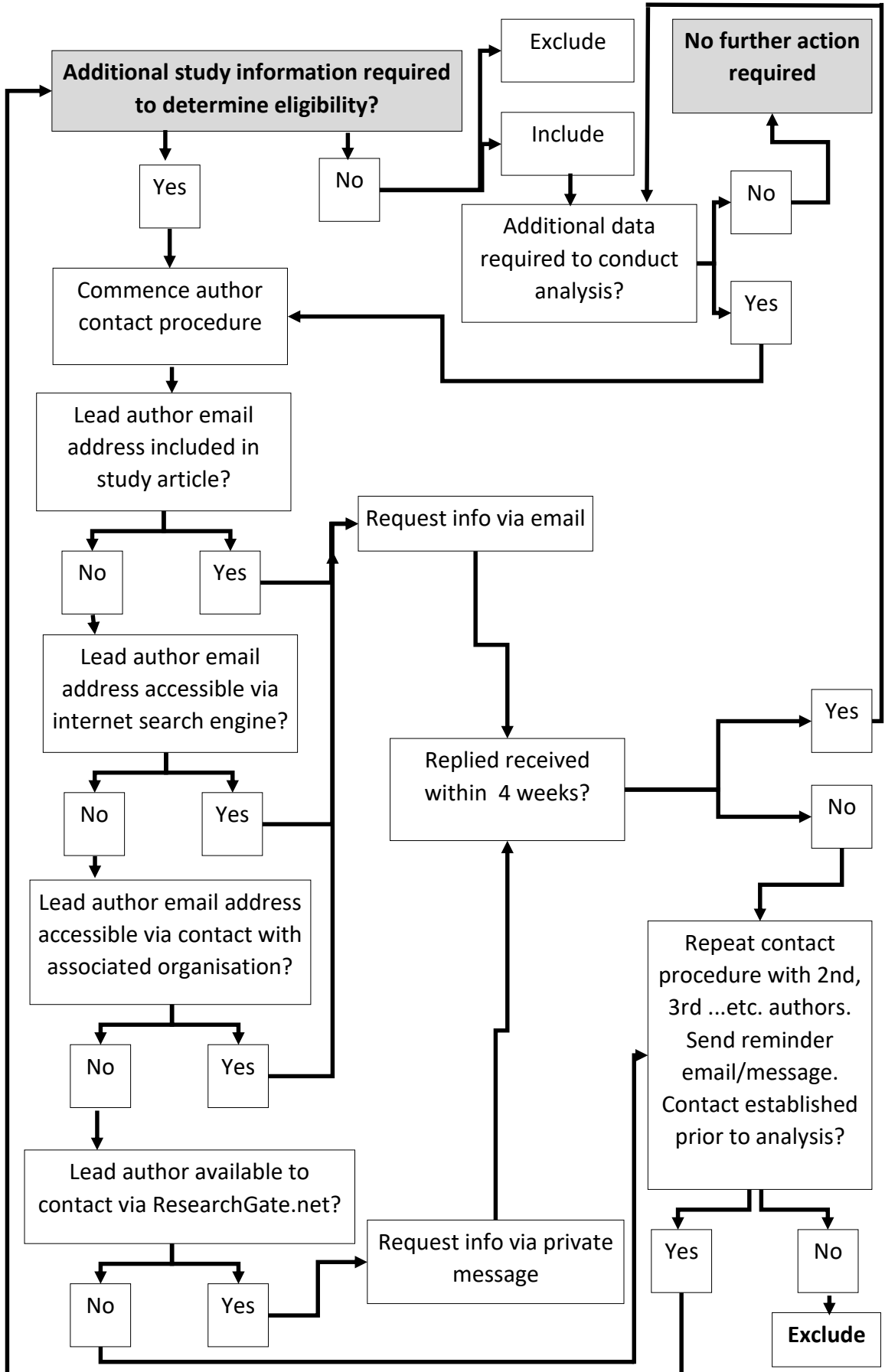
(d) Trial OR trials OR randomi*ed OR RCT OR "pre and post" OR effectiveness OR effect OR efficacy OR quantitative OR outcome OR outcomes OR comparison

Adaptations for each source:

Source	Terms	Limits
PsycINFO	As above	English Language; Adulthood 18+; Clinical Trial; Empirical Study; Field Study; Quantitative study; Treatment Outcome
MEDLINE	As above	English Language; All Adult; Human; Clinical Trial; Comparative Study; Controlled Clinical Trial; Journal Article;
CINAHL	As above	English Language; All Adult; Clinical Trial; Doctoral Dissertation; Journal Article; Masters Thesis; Nursing Interventions; Randomized Controlled Trial; Research
Scopus	As above	Article; Article in Press; Article or Conference Paper
Open Grey	As above	None
ProQuest	As above	None
ISRCTN registry	As above	None
ClinicalTrials.gov	(a) AND (b) AND (Therapy OR CBT OR ACT OR MBCT OR DBT OR "behavio*arl activation" OR "systematic desensiti*ation")	Adult; Senior

Appendix F

Procedure for Contacting Study Authors



Appendix G

Representation of Electronic Adapted RCT Psychotherapy Quality Rating Scale

Adapted RCT of Psychotherapy Quality Rating Scale

Instructions: Complete all applicable orange shaded boxes. If not applicable leave shaded box blank, but enter "N/A ...incl. reason" in cell adjacent. Subtotals and final total are calculated automatically (shaded blue).

Study ID:

Item	<i>Description of Participants</i>	Highlight
1	Diagnostic method and criteria for inclusion and exclusion (<u>for person with dementia</u>)	0 poor description (e.g. just uses umbrella term "has dementia" / "memory problems") AND inappropriate method/criteria (e.g. no mention of diagnostic screening or clinical diagnosis)
		1 full description (describes/reports dementia type, e.g. "any progressive dementia", "Alzheimer's" etc.) OR appropriate method/criteria (PwD met diagnostic manual criteria, Carer reported PwD had clinical diagnosis).
		2 full description (describes/reports dementia type, e.g. "any progressive dementia", "Alzheimer's" etc.,) AND appropriate method/criteria (PwD met diagnostic manual criteria, Carer reported PwD had clinical diagnosis).
2	Identification method and criteria for inclusion and exclusion (<u>carer</u>)	0 poor description (just states "carer") and inappropriate method/criteria (e.g. self-selected, no or limited use of criteria to define the carer)
		1 full description (includes caregiving details e.g. relationship to PwD, time spent caregiving etc., exclusion criteria) OR appropriate method/criteria (interview, screening questionnaire, psychometrics).
		2 full description (includes caregiving details, e.g. relationship to PwD, time spent caregiving etc., exclusion criteria) AND appropriate method/criteria (interview, screening questionnaire).

3	Documentation or demonstration of reliability of Participant selection method	0	poor or no reliability documentation
1		brief reliability documentation (sufficient, even if it is not explicitly cited). e.g. May state inter-rater reliability of Pp selection was ensured, but have not reported values OR may have used known reliable methods e.g. widely known reliable psychometric measure (e.g. BDI) but not explicitly commented on reliability.	
2		full reliability documentation (within-study reliability necessary). Inter-rater reliability of participant selection is tested and reported AND If psychometric measures are used in the selection criteria, the reliability of the measure must be referred to.	
4	Description of relevant comorbidities in carer	0	poor or no description
1		brief description of relevant comorbidities in carer. (Either briefly states comorbidities which would/would not result in exclusion. This may be a brief description of several comorbidities e.g. "Pps with other conditions were excluded", OR briefly describes comorbidities in sample).	
2		full description of relevant comorbidities in carer (states comorbidities which would/would not result in exclusion OR describes any comorbidities -or comments on lack of- in the sample; e.g. psychosis, brain injury, any other MH condition)	
5	Description of numbers of participants screened, included and excluded	0	poor or no description
1		brief description (e.g. states numbers included and excluded OR qualitative information detailing reasons for exclusion)	
2		full description (includes numbers included and excluded AND includes reasons for exclusion. May be presented in text or as flowchart.)	
<i>Definition and Delivery of Intervention</i>			
6	Interventions (including control/comparison groups) are sufficiently	0	poor or no description or references for control group AND intervention group (e.g. much info may have had to be obtained from author to establish study eligibility)

described or referenced to allow for replication	1	brief intervention description or references for intervention group AND control group (may give a brief outline or reference for intervention and control aims but not enough to enable complete replication). OR full description or references of one group (with brief/poor description of the other group)
	2	full intervention description or references (manual not required) for BOTH intervention and control group (<u>For intervention</u> , may reference and cite a therapeutic programme on which the intervention group is based/psychological theory underpinning the intervention AND will include a description of the intervention plan, such as the content of each session, or have made reference to where to locate such information. <u>For control group</u> may state the intention of the control group, e.g. to act as 'waitlist control' / 'treatment as usual', "not intended to challenge thoughts" etc. AND describe the control group content.
7	0	Poor or no adherence reporting
Method to demonstrate that intervention being studied is intervention being delivered (only satisfied if transcripts or tapes are explicitly reviewed, or if observed by supervisor). Only rate for applicable interventions	1	Brief adherence reporting (Evidence of intervention facilitator having supervision (e.g. individual/group supervision) OR adherence to intervention assessed using a measure e.g. CBT CTSR, or self-report measure.
	2	Full adherence reporting (Evidence of intervention facilitator having supervision (e.g. individual/group supervision) AND adherence to intervention assessed using a measure e.g. CBT CTSR, but not self-report measure, must be completed by another person)
8	0	no description of therapist training/experience partial description of therapist training/experience (Job role OR qualification level / accreditation OR level at which the intervention will be delivered, e.g. 'suitable for health care assistants' if different to facilitator qualifications)
Appropriate description of therapist training and level of experience in the intervention under investigation (where applicable)	1	full description of therapist training (Job role AND qualification level / accreditation AND level at which the intervention will be delivered, e.g. 'suitable for health care assistants' <u>if</u> different to facilitator qualifications)
	2	full description of therapist training (Job role AND qualification level / accreditation AND level at which the intervention will be delivered, e.g. 'suitable for health care assistants' <u>if</u> different to facilitator qualifications)

9	Therapist supervision while treatment is being provided as appropriate to intervention (where applicable)	0	poor description and inadequate therapist supervision (where supervision would be required, i.e. supervision not necessary for self-help interventions)
		1	full description OR adequate therapist supervision as appropriate to intervention
		2	full description AND adequate therapist supervision as appropriate to intervention (description includes factors such as frequency/duration and profession of supervisor. Supervision is adequate if appropriate to intervention, e.g. for short term CBT, therapist to be supervised by a senior/experienced colleague, for self-help intervention supervision not required for rating of 2)
10	Description of concurrent interventions (e.g. medication) allowed and administered during the course of study (if patients on medications are included, a rating of 2 required full reporting of what medications were used; if patients on medications are excluded, this alone is sufficient for a rating of 2)	0	poor or no description of concurrent treatments
		1	brief description of concurrent treatments (e.g. participants continued to take any previously prescribed medication, participants to continued attending other support) OR brief description of concurrent treatments which would result in exclusion (e.g. excluded if accessing other support)
		2	full description of concurrent treatments (e.g. participants already taking anti-depressant medication continued to do so during the study, participants continued attending social support groups, participants taking medication excluded) OR full description of concurrent treatments which would result in exclusion (e.g. excluded if currently engaging in CBT/ other self-help programme etc.)
<i>Outcome Measures</i>			
11	Validated outcome measures (either established or newly established)	0	poor or no validation of outcome measures
		1	brief validation of ALL outcome measures (shown or cited) OR full validation of SOME outcome measures
		2	full validation of ALL outcome measures (shown or cited)

12	Primary outcome measures specified in advance (although does not need to be stated explicitly for a rating of 2)	0	poor specification in advance (e.g. may not specify which outcomes are primary (however note, if all outcomes are given equal importance wouldn't expect primary/secondary to be stated), OR may state outcome X is primary, but discuss outcome Y in more detail and neglect outcome X in discussion.
		1	brief specification in advance (e.g. some acknowledgement of which outcomes are primary e.g. a measure of burden, and which are secondary)
		2	full specification in advance (e.g. the Zarit Burden questionnaire is the primary outcome, BDI depression will be examined as a secondary outcome), it can be helpful to check that primary outcomes are treated as such in the discussion, to ensure the researcher has not changed the primary focus after obtaining the results.
13	Outcome assessment by rater blinded to intervention group and with establish reliability	0	poor or no blinding (e.g. rating by therapist who is aware of intervention group) OR poor information and unable to determine
		1	blinding of independent rater to treatment group OR established reliability (inter-rater reliability completed)
		2	blinding of independent rater to treatment group AND established reliability (inter-rater reliability completed) . Note, inter rater reliability not required for self-report scales, but must have been 'handed-out' to participants by an independent person who was not involved in the intervention.
14	Discussion of safety and adverse events during study intervention	0	poor or no discussion
		1	brief details (e.g. PwD death, brief details re negative effects of intervention)
		2	details and full discussion (e.g. as above but with full details and discussion. E.g. x3 PwD deaths due to dementia. Safety discussion may include e.g. if scored above a cut-off, or presented as highly distressed, referred for help)
15	Assessment of long-term post-termination	0	poor or no post-termination assessment of outcome

	outcome (should not be penalised for failure to follow comparison group if this is a wait-list or non-treatment group that is subsequently referred for active intervention)	1	medium-term assessment of outcome (2 - 12 months post-termination)
		2	long-term assessment of outcome (≥ 12 months post-termination)
16	Inclusion of Social Significance and Social Validity outcomes as recommended by Schulz et al. 2002	0	No measures of social validity or social significance included
		1	Measure or report of either Social Significance (e.g. rate of PwD admission to residential care) OR Social Validity (e.g. acceptability of the intervention, ease of completion) Can be quantitative measure or qualitative description. Measure or report of Social Significance (e.g. quality of life, rate of PwD admission to residential care) AND Social Validity (e.g. acceptability of the intervention, ease of completion)
<i>Data Analysis</i>			
17	Intent-to-treat method for data analysis involving primary outcome measure	0	no description or no intent-to-treat analysis with primary outcome measure
		1	partial intent-to-treat analysis (excludes participants with no follow-up data) with primary outcome measure
		2	full intent-to-treat analysis (included outcome data for all participants) with primary outcome measure
18	Description of dropouts and withdrawals	0	poor or no description
		1	brief description (may include numbers OR reasons)
		2	full description (numbers must be explicitly stated AND include reasons for drop outs and withdrawals)
19	Appropriate statistical test (e.g. use of Bonferroni correction, longitudinal data analysis, adjustment only for a priori identified confounders)	0	inappropriate statistics, extensive data dredging, or no information about appropriateness of statistics
		1	moderately appropriate, though unsophisticated, statistics and/or moderate data dredging
		2	fully appropriate statistics and minimal data dredging in primary findings
20	Adequate sample size	0	inadequate justification AND inadequate sample size

		1	adequate justification OR adequate sample size (based on guidelines for RCT)
		2	adequate justification AND adequate sample size (e.g. a priori sample size calculation and sample size achieved)
21		0	not discussed or considered
	Appropriate consideration of therapist (where intervention is therapist facilitated) and site effects	1	discussed OR considered statistically (e.g. cluster analysis, random effects, marginal models)
		2	discussed AND considered statistically (e.g. cluster analysis, random effects, marginal models)
<i>Intervention Assignment</i>			
22	A priori relevant hypotheses that justify comparison group	0	poor or no justification
		1	brief or incomplete justification
		2	full justification of comparison group
23		0	comparison group from significantly different population and/or time frame
	Comparison group from same population and time frame as experimental group	1	from moderately different population/time frame OR states from the same population/time frame (but have not demonstrated statistically)
		2	from the same population/time frame (incl. stats analysis on demographics to show no differences between groups at baseline, OR analysis shows any differences in characteristics do not affect outcome)
24		0	poor (e.g. pseudo-randomisation, sequential assignment) or no randomisation
	Randomised assignment to intervention groups	1	adequate but poorly defined randomisation procedure
		2	full and appropriate method of randomisation performed after screening and baseline assessment (method must be described, e.g. XXX software)
25		0	no information OR poor balance of allegiance by study therapists (e.g. therapy in experimental and control groups both administered by therapists with strong allegiance to therapy being tested in the experimental group)
	Balance of allegiance to types of intervention by practitioner (where applicable, not applicable for self-help)	1	some balance if allegiance to interventions by study therapists OR limited information (e.g. may be info about therapist in intervention group but not control group)

- 2 full balance of allegiance to interventions (e.g. therapies administered by therapists with allegiance to respective techniques)

Overall Quality of Study

26	Conclusions of study justified by sample, measures, and data analysis, as presented (note: useful to look at conclusions as stated in study abstract)	0	Poor or no justification of conclusions from results as presented OR insufficient information to evaluate (e.g. sample or intervention insufficiently documented, data analysis does not support conclusions, or numbers of withdrawals or dropouts make finding unsupportable)
		1	some conclusion of study justified OR partial information presented to evaluate
		2	all conclusions of study justified AND complete information presented

Number of Applicable items in questions 1 to 26? **Please ensure correct count**

Subtotal for Items 1 to 26

Percentage score (use to inform item 26 Omnibus rating)

27	Omnibus rating: please provide an overall rating of the quality of the study, taking into account the adequacy of description, the quality of study design, data analysis and justification of conclusions	1	exceptionally poor ($\leq 15\%$)
		2	very poor (16 to 29 %)
		3	moderately poor (30 to 44%)
		4	average (45 to 60%)
		5	moderately good (60 to 72%)
		6	very good (73 to 88%)
		7	exceptionally good (88 to 100%)

**Total Adapted RCT of
Psychotherapy Quality
Rating Scale Score**

###

**Total Adapted RCT of
Psychotherapy Quality
Rating Scale Score
Percentage**

###

%

Appendix H

Representation of Electronic Data Extraction Form for Empirical Paper Two

Instructions: Complete all items on the form. Where multiple choice, highlight option (including number) using BOLD format. Where item states "specify", type text answer in adjacent cell. Use 'Additional Info' column for relevant comments, e.g. if have contacted an author to obtain more info.

Study ID:

	Authors	Specify:
	Publication date	Specify:
	Source	1 Peer reviewed journal 2 Journal 3 Abstract 4 Conference abstract 5 Contact with author 6 Theses & dissertations 7 Other (specify)
Study	Study Design	1 RCT 2 CCT 3 Pragmatic Trial 4 Other (specify)
	Method of allocating participants to groups	1 Randomised 2 Quasi-randomised 3 Non-randomised 4 Other (specify)
	Country	Specify:
	Language	Specify:
	Ethnicity	Specify: (Majority & minorities)
Participants	Relationship to the care recipient	1 Spouse 2 Adult child 3 Sibling 4 Parent 5 Friend 6 Combination of above 7 Other (specify)

Presence of psychiatric diagnosis in carer	<ol style="list-style-type: none"> 1 Depression 2 Anxiety 3 Other (specify) 4 No diagnosis
Diagnosis tool (e.g. DSM, ICD)	<ol style="list-style-type: none"> 1 DSM IV 2 DSM V 3 ICD-10 4 N/A - no diagnosis 5 Other (specify)
Cared for person's dementia diagnosis	<ol style="list-style-type: none"> 1 Alzheimer's 2 Vascular 3 Lewy body 4 Fronto-temporal 5 Combination of above (e.g. any dementia) 6 Other (specify)
Total Participants	Number: Mean Age: Standard Deviation of Age: Range of Age: Gender - percentage female %: Mean number of hours spent caring: Standard deviation of hours spent caring p/w: Mean number of number of months as caregiver: Standard deviation of number of months as caregiver:
Completer Participants (Combined Intervention & Control group)	Number: Mean Age: Standard Deviation of Age: Range of Age: Gender - percentage female %: Mean number of hours spent caring p/w: Standard deviation of hours spent caring: Mean number of number of months as caregiver: Standard deviation of number of months as caregiver:
Dropout Participants (Combined Intervention & Control group)	Number:

		<p>Mean Age:</p> <p>Standard Deviation of Age:</p> <p>Range of Age:</p> <p>Gender - percentage female %:</p> <p>Mean number of hours spent caring:</p> <p>Standard deviation of hours spent caring p/w:</p> <p>Mean number of number of months as caregiver:</p> <p>Standard deviation of number of months as caregiver:</p>
	Intervention Group Participants	<p>Number:</p> <p>Mean Age:</p> <p>Standard Deviation of Age:</p> <p>Range of Age:</p> <p>Gender - percentage female %:</p> <p>Mean number of hours spent caring:</p> <p>Standard deviation of hours spent caring p/w:</p> <p>Mean number of number of months as caregiver:</p> <p>Standard deviation of number of months as caregiver:</p>
	Control Group Participants	<p>Number:</p> <p>Mean Age:</p> <p>Standard Deviation of Age:</p> <p>Range of Age:</p> <p>Gender - percentage female %:</p> <p>Mean number of hours spent caring p/w:</p> <p>Standard deviation of hours spent caring:</p> <p>Mean number of number of months as caregiver:</p> <p>Standard deviation of number of months as caregiver:</p>
Intervention	Format	<p>1 Talking therapy</p> <p>2 Bibliotherapy</p> <p>3 Computerised</p> <p>4 Telephone</p> <p>5 Combined (select all that apply)</p> <p>6 Other (specify)</p>
	Individual / group	<p>1 Individual</p> <p>2 Group</p> <p>3 Combined individual and group</p> <p>4 Other (specify)</p>
	Facilitation	<p>1 Supported (specify total number of hours contact time)</p>

	2	Unsupported
Number of sessions		Specify:
Session Duration		Specify:
Facilitator Qualification	1	No relevant formal qualification or training (e.g. HCA, NA)
	2	Academic bachelors degree level qualification (e.g. Psychology BSc degree)
	3	Academic Masters degree level qualification (e.g. MSc Psychology) but NOT accredited/licensed clinical qualification
	4	Academic Doctoral level qualification (e.g. Psychology research doctorate, nursing research doctorate) NOT able to engage in independent clinical practice i.e. deliver therapy with a clinical population
	5	Professional Clinical Bachelors level qualification (e.g. Registered Mental Health Nurse, OT, Social Worker)
	6	Professional Clinical Masters degree level qualification (e.g. MSc CBT), must be accredited/licensed to practice independently
	7	Professional Clinical Doctorate level qualification (e.g. Clinical Psychologist, Counselling Psychologist) must be accredited/licensed to practice independently
	8	Multiple facilitators (highlight all that apply)
	9	Other (specify)
	10	N/A - not facilitated
Facilitator Profession	1	Non-professional
	2	Nursing (incl. RMN, Nursing Assistant)
	3	Social Work
	4	Occupational Therapy
	5	Counselling
	6	Psychology / Psychological Therapy
	7	Mental Health Worker
	8	Researcher
	9	multiple facilitators (highlight all that apply)
	10	Other (specify)
	11	N/A - not facilitated
Facilitator Given Training specific to Studied Intervention?	1	Yes (Specify total hours training time)
	2	No
	3	N/A - not facilitated

	Approach	<p>CBT based only (all aspects of intervention based on CBT and includes use of CBT therapeutic techniques)</p> <p>CBT psychoeducation only (does not include engaging in therapy, purely psychoeducational)</p> <p>CBT based multi-component (includes CBT based components but may also include other components not based on CBT)</p> <p>3rd wave CBT based (e.g. ACT, CFT; Specify)</p> <p>Other (specify)</p>
	Presence of cared for person during intervention	<p>Present</p> <p>Partially present</p> <p>Not present</p> <p>Not known</p>
Comparison	Control Group	<p>Placebo</p> <p>Waitlist</p> <p>Treatment as usual</p> <p>No treatment</p> <p>Other non-active control (specify)</p>
	Measurement Time Points	<p>Pre & Post</p> <p>Pre, Post & x1 Follow-up (Specify follow-up time point, e.g. 3 months)</p> <p>Pre, Post & Multiple Follow-up (Specify time points, e.g. 3 months & 1 year)</p> <p>Other (specify)</p>
Outcomes	Areas of Outcome	<p>Depression</p> <p>Anxiety</p> <p>Burden</p> <p>Distress (Stress/Strain)</p> <p>Measure of social significance (e.g. admission of cared for person to a care home)</p> <p>Measure of social validity (e.g. acceptability of intervention)</p> <p>Other (specify)</p> <p>Other (specify)</p> <p>Other (specify)</p>
	Depression	<p>Measure used:</p> <p>Measure validity (incl. details):</p> <p>Measure reliability (incl. details):</p> <p>N/A Depression not an outcome</p>
	Depression unit of measurement	<p>Dichotomous</p> <p>Categorical</p>

		3 Continuous
		4 N/A Depression not an outcome
	Anxiety	1 Measure used: 2 Measure validity (incl. details): 3 Measure reliability (incl. details): 4 N/A Anxiety not an outcome
	Anxiety unit of measurement	1 Dichotomous 2 Categorical 3 Continuous 4 N/A Anxiety not an outcome
	Burden	1 Measure used: 2 Measure validity (incl. details): 3 Measure reliability (incl. details): 4 N/A Burden not an outcome
	Burden unit of measurement	1 Dichotomous 2 Categorical 3 Continuous 4 N/A Burden not an outcome
	Distress (Stress/strain)	1 Measure used: 2 Measure validity (incl. details): 3 Measure reliability (incl. details): 4 N/A stress/strain not an outcome
	Stress/Strain unit of measurement	1 Dichotomous 2 Categorical 3 Continuous 4 N/A Stress/strain not an outcome
Results	Participants	Intervention group N at baseline Intervention group N at post measure Intervention group N at follow-up (if applicable) Number drop-outs from baseline to post Baseline to post attrition rate % Number drop-outs from baseline to follow-up (if applicable) Baseline to follow-up attrition rate % (if applicable)
		Identified reasons for Drop-out (If multiple, record all reasons with percentage in brackets, <i>e.g. Pp death(2), not stated (3)</i>)
		Control group N at baseline Control group N at post measure Control group N at follow-up (if applicable) Number drop-outs from baseline to post

	Baseline to post attrition rate %
	Number drop-outs from baseline to follow-up (if applicable)
	Baseline to follow-up attrition rate % (if applicable)
	Identified reasons for Drop-out (If multiple, record all reasons with percentage in brackets, e.g. Pp death(2), not stated (3))
	Depression Outcome measure used:
	Primary Analysis method
	Follow-up Analysis method
	Intervention group Mean - Baseline
	Intervention group SD - Baseline
	Intervention group Mean - Post
	Intervention group SD - Post
	Intervention group Mean - Follow-up (if applicable)
	Intervention group SD - Follow-up (if applicable)
	Effects size type measured
	Effects size value Baseline to Post
	Effects size value Baseline to Follow-up (if applicable)
	Control group Mean - Baseline
	Control group SD - Baseline
	Control group Mean - Post
	Control group SD - Post
	Control group Mean - Follow-up (if applicable)
	Control group SD - Follow-up (if applicable)
	Effects size type measured
	Effects size value Baseline to Post
	Effects size value Baseline to Follow-up (if applicable)
	Reported statistical outcome (e.g. $t=...$, $p=...$)
	Anxiety Outcome measure used:
	Primary Analysis method
	Follow-up Analysis method
	Intervention group Mean - Baseline
	Intervention group SD - Baseline
	Intervention group Mean - Post
	Intervention group SD - Post
	Intervention group Mean - Follow-up (if applicable)
	Intervention group SD - Follow-up (if applicable)
	Effects size type measured
	Effects size value Baseline to Post

Depression

Anxiety

	Effects size value Baseline to Follow-up (if applicable)
	Control group Mean - Baseline
	Control group SD - Baseline
	Control group Mean - Post
	Control group SD - Post
	Control group Mean - Follow-up (if applicable)
	Control group SD - Follow-up (if applicable)
	Effects size type measured
	Effects size value Baseline to Post
	Effects size value Baseline to Follow-up (if applicable)
	Reported statistical outcome (e.g. $t=...$, $p=...$)
	Burden Outcome measure used:
	Primary Analysis method
	Follow-up Analysis method
	Intervention group Mean - Baseline
	Intervention group SD - Baseline
	Intervention group Mean - Post
	Intervention group SD - Post
	Intervention group Mean - Follow-up (if applicable)
	Intervention group SD - Follow-up (if applicable)
	Effects size type measured
	Effects size value Baseline to Post
	Effects size value Baseline to Follow-up (if applicable)
	Control group Mean - Baseline
	Control group SD - Baseline
	Control group Mean - Post
	Control group SD - Post
	Control group Mean - Follow-up (if applicable)
	Control group SD - Follow-up (if applicable)
	Effects size type measured
	Effects size value Baseline to Post
	Effects size value Baseline to Follow-up (if applicable)
	Reported statistical outcome (e.g. $t=...$, $p=...$)
	Distress (Stress/Strain) Outcome measure used:
	Primary Analysis method
	Follow-up Analysis method
	Intervention group Mean - Baseline
	Intervention group SD - Baseline
	Intervention group Mean - Post
Burden	
Distress (Stress/Strain)	

Intervention group SD - Post

Intervention group Mean - Follow-up (if applicable)

Intervention group SD - Follow-up (if applicable)

Effects size type measured

Effects size value Baseline to Post

Effects size value Baseline to Follow-up (if applicable)

Control group Mean - Baseline

Control group SD - Baseline

Control group Mean - Post

Control group SD - Post

Control group Mean - Follow-up (if applicable)

Control group SD - Follow-up (if applicable)

Effects size type measured

Effects size value Baseline to Post

Effects size value Baseline to Follow-up (if applicable)

Reported statistical outcome (e.g. $t=...$, $p=...$)

Appendix I

Overview of Intervention Groups and Control Groups for Studies Included in the Second Empirical Paper

Study	Intervention overview	Control type and Overview
Au et al. (2010)	'Coping with Caregiving' comprising a variety of cognitive and behavioural skills and education about dementia caregiving	Waitlist
Beauchamp et al. (2005)	Caregiver's Friend: Dealing with Dementia', worksite web-based intervention, including knowledge, cognitive and behavioural skills, and affective learning	Waitlist
Blom et al. (2015)	Mastery over Dementia' internet intervention, including problem solving, cognitive restructuring, assertiveness training and relaxation	Non-active e-bulletins with practical information e.g. holiday breaks
Chang (1999)	CBT for CG and cared for person dyads tailored to dressing and eating difficulties	Non-active attention control
Chiu et al. (2015)	Problem solving techniques	Treatment as usual
Dowling et al (2013)	Life Enhancing Activities for Family Caregivers' using cognitive and behavioural skills to increase positive affect, as well as mindfulness and altruistic behaviours	Non-active attention control
Gallagher-Thompson et al. (2008a)	Coping with Caregiving' comprising a variety of cognitive and behavioural skills and education about dementia caregiving	Non-active empathic telephone calls and postal information about dementia
Gallagher-Thompson et al. (2008b)	Coping with Caregiving' comprising a variety of cognitive and behavioural skills and education about dementia caregiving	Non-active empathic telephone calls and postal information about dementia
Kajiyama et al. (2013)	iCare Stress Management' including cognitive and behavioural techniques, as well as information about dementia, behaviour management and communication skills	Non-active generic education about dementia
Steffen (2000)*	Anger management intervention including tension-reduction strategies, cognitive change strategies and assertion training	Waitlist

Steffen and Grant (2016)	Telehealth behavioural coaching' including behavioural activation, behaviour management and relaxation	Treatment as usual
Tremont et al. (2008)	Family Intervention: Telephone Tracking - Dementia' (FITT-D), including active problem solving and facilitating positive changes within the family system	Treatment as usual
Villareal-Reyna et al. (2012)	Cognitive Conduct' including changing dysfunctional thoughts, cognitive-behavioural mood management, and humour/laughter	Non-active 'home accident prevention' information group

Note. *Composite study combining Steffen 2000a & 2000b; **Steffen 2000a; ***Steffen 2000b; NR = Not reported.

Appendix J

Quality Rating Sub-total Scores for Studies Included in the Second Empirical Paper

Study	Description of Participants (max = 10)	Definition and Delivery of Intervention (max = 10)	Outcome Measures			Data Analysis (max = 10)	Intervention Assignment (max = 8)	Overall Quality of Study Conclusions (max = 2)	Total Quality Rating (%)
			Inclusion of Social Significance	Inclusion of Social Validity	Outcome Measures Total (max = 12)				
Au et al. (2010)	5	5	No	No	4	2	4	1	Moderately poor (40.7)
Beauchamp et al. (2005)	2	2	No	Yes	3	3	4	1	Moderately poor (34.7)
Blom et al. (2015)	7	3	No	No	7	7	5	1	Average (57.6)
Chang (1999)	2	1	No	No	4	4	4	2	Moderately poor (33.9)
Chiu et al. (2015)	2	3	No	No	7	6	6	2	Average (50.8)
Dowling et al. (2013)	4	3	No	Yes	5	2	4	1	Moderately poor (35.6)
Gallagher-Thompson et al. (2008a)	6	6	No	No	6	5	4	1	Average (54.2)

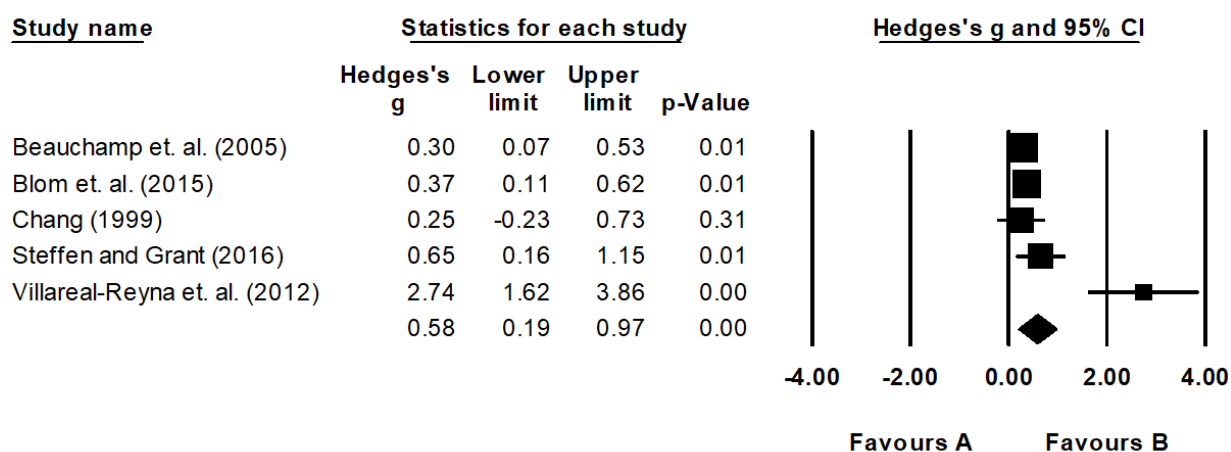
Gallagher-Thompson et al. (2008b)	6	6	No	No	6	5	4	1	Average (54.2)
Kajiyama et al. (2013)	8	2	No	Yes	5	5	5	2	Moderately good (63.3)
Steffen (2000)*	5	8	No	Yes	3	3	4	1	Average (45.8)
Steffen and Grant (2016)	8	4	Yes	No	8	5	7	1	Moderately good (62.7)
Tremont et al. (2008)	7	6	No	Yes	8	3	5	1	Average (55.9)
Villareal-Reyna et al. (2012)	9	6	No	No	4	6	2	1	Average (54.2)

Appendix K

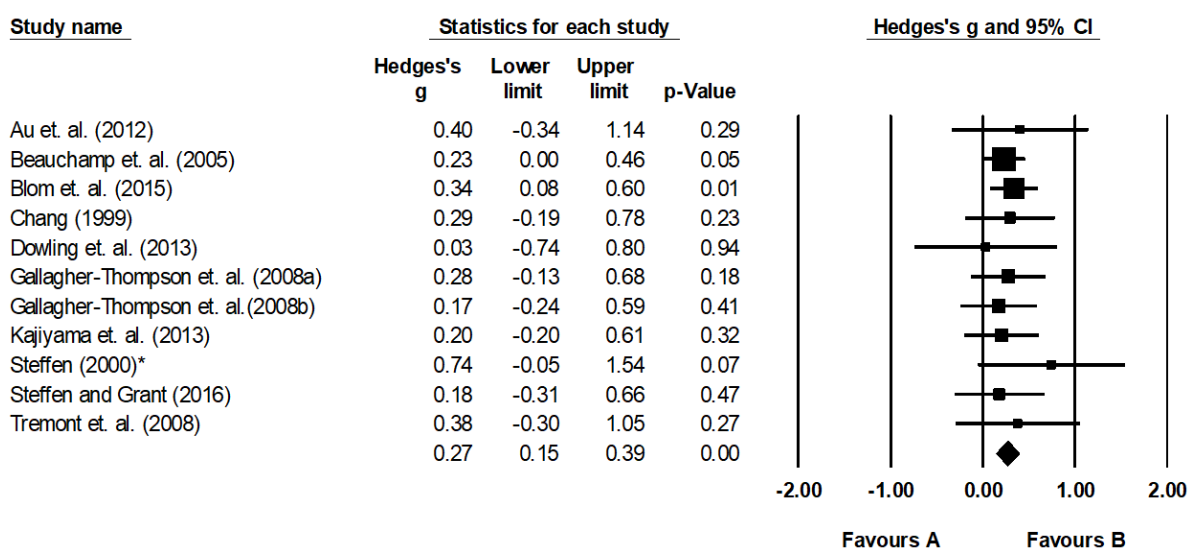
Forest Plots for Meta-analyses of Anxiety, Depression, Burden and Distress Outcomes Conducted in the Second Empirical Paper

Note. Favours A = favours control group; Favours B = favours intervention group.

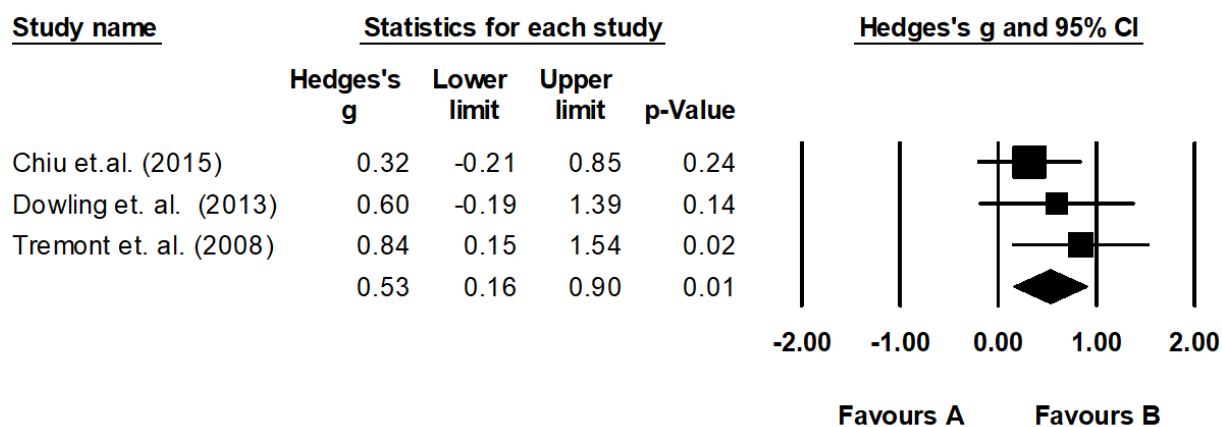
Anxiety Meta Analysis



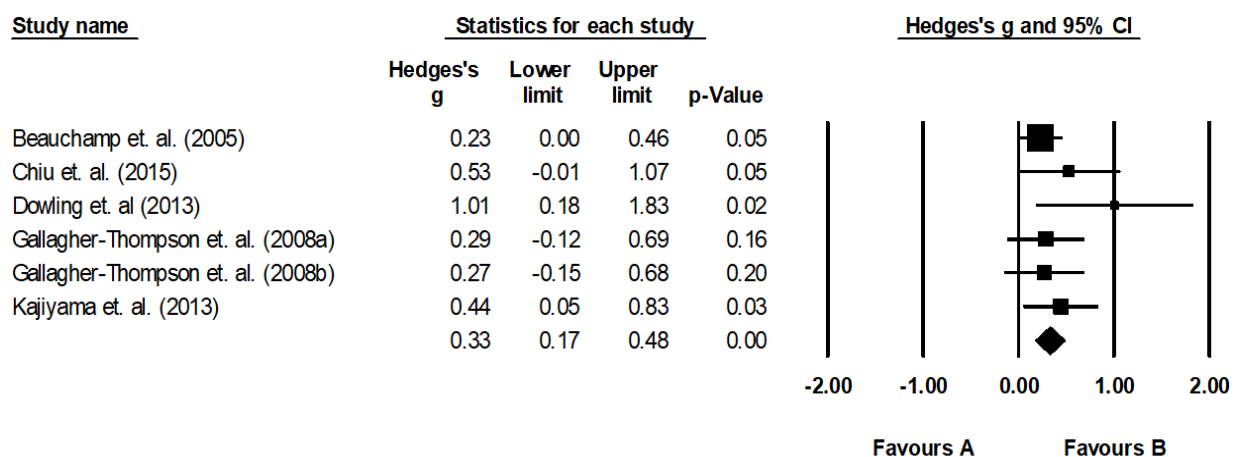
Depression Meta Analysis



Burden Meta Analysis



Distress Meta Analysis



Appendix L

Publication Bias Funnel Plots for Anxiety, Depression, Burden and Distress Meta-Analyses in the Second Empirical Paper

Note. Displays observed (outlined) and imputed studies (filled)

