

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

Laura Kaddour

Department of Clinical Psychology, Norwich Medical School, University of East Anglia,
Norwich, UK

Dr Naoko Kishita

School of Health Sciences, University of East Anglia, Norwich, UK

Anthony Schaller

UK

Correspondence concerning this article should be addressed to Laura Kaddour, Department
of Clinical Psychology, Norwich Medical School, University of East Anglia, Norwich, NR4
7TJ, UK. Email: L.Kaddour@uea.ac.uk.

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Design: Literature searches were conducted in databases of published (PsycINFO; MEDLINE; CINAHL; Scopus) and unpublished (Open Grey; ISRCTN registry; ClinicalTrials.gov; ProQuest) literature.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 dementia 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 & 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 relevant outcomes (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 (see appendix A1 published as supplementary material online attached to the electronic version

of this paper at <https://www.cambridge.org/core/journals/international-psychogeriatrics>).

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 progressive dementia illness. (b) The intervention was based on CBT theory or techniques (Beck, 1979). For example, problem solving, thought challenging, behavioural activation, graded exposure etc. (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 accredited/licensed and non-accredited/licensed facilitators 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. Missing information related to intervention intensity and frequently included facilitator profession/qualification level. Articles were subsequently excluded if additional information was not provided prior to data extraction (Figure 1).

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, 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 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 had missing data, two of which were subsequently excluded due to being unable to obtain data from authors (Figure 1).

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 1. 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 1. 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 of people with any dementia or Alzheimer's disease. All samples were from developed countries, based on the Human Development Index (United Nations Development Programme, 2016).

Intervention characteristics are summarised in Table 2. The majority of studies delivered multi-component CBT-based interventions, combining CBT techniques with other non-CBT techniques (e.g. 'identifying local resources' education) (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 Table 3.

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).

Study Quality and Risk of Bias

The quality of included studies presented in Table 4 ranged from 'moderately poor' to 'moderately good'. Four studies were rated as 'moderately poor' (Chang, 1999; Beauchamp *et al.*, 2005; Au *et al.*, 2010; Dowling *et al.*, 2013), eight were rated as 'average' (Steffen 2000a; 2000b; Gallagher-Thompson *et al.*, 2008a; 2008b; Tremont *et al.*, 2008; Villareal-Reyna *et al.*, 2012; Blom *et al.*, 2015; Chiu *et al.*, 2015), and two were rated as 'moderately good' (Kajiyama *et al.*, 2013; Steffen and Grant, 2016).

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. Three studies reported follow-up data at one-month post intervention completion (Chang, 1999; Villareal-Reyna *et al.*, 2012; Dowling *et al.*, 2013), two reported two-month follow-up outcomes (Gallagher-Thompson *et al.*, 2008a; 2008b) and only one reported six-month follow-up data (Steffen and Grant, 2016). 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, $p<0.01$) (see appendix A2 published as supplementary material online attached to the electronic version of this paper at <https://www.cambridge.org/core/journals/international-psychogeriatrics>). 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 (see appendix A3 published as supplementary material online attached to the electronic version of this paper at <https://www.cambridge.org/core/journals/international-psychogeriatrics>). 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, $p < 0.01$) (see appendix A2 published as supplementary material online attached to the electronic version of this paper at <https://www.cambridge.org/core/journals/international-psychogeriatrics>). 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, $p = 0.01$) (see appendix A2 published as supplementary material online attached to the electronic version of this paper at <https://www.cambridge.org/core/journals/international-psychogeriatrics>). 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 (see appendix A3 published as supplementary material online attached to the electronic version of this paper at <https://www.cambridge.org/core/journals/international-psychogeriatrics>). 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, $p<0.01$) (see appendix A2 published as supplementary material online attached to the electronic version of this paper at <https://www.cambridge.org/core/journals/international-psychogeriatrics>). 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 (see appendix A3 published as supplementary material online attached to the electronic version of this paper at <https://www.cambridge.org/core/journals/international-psychogeriatrics>). 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 intense. 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 small number of studies was unexpected given that distress is commonly reported on in the DC literature. However, 'distress' is often used an umbrella term to refer to the other variables examined in this paper

(i.e. anxiety, depression and burden) (Ferrara *et al.*, 2008; Cress, 2009). Therefore, labelling outcomes as 'distress' should be avoided to prevent ambiguity, and the use of this term can be considered a limitation of this paper.

Nonetheless, it is recommended that future DC intervention studies should include stress/strain as an outcome, particularly as it has been suggested that stress/strain may mediate psychological morbidity (Schulz and Martire, 2004).

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. 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.

Conflicts of Interest

None.

Description of Author Roles

L. Kaddour designed the study, extracted and quality assessed data, conducted statistical analysis and wrote the article. A. Schaller extracted and quality assessed data, and resolved discrepancies with L. Kaddour. Dr N. Kishita supervised the study design, data extraction and the writing of the article.

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Figure 1. Systematic literature search flow chart

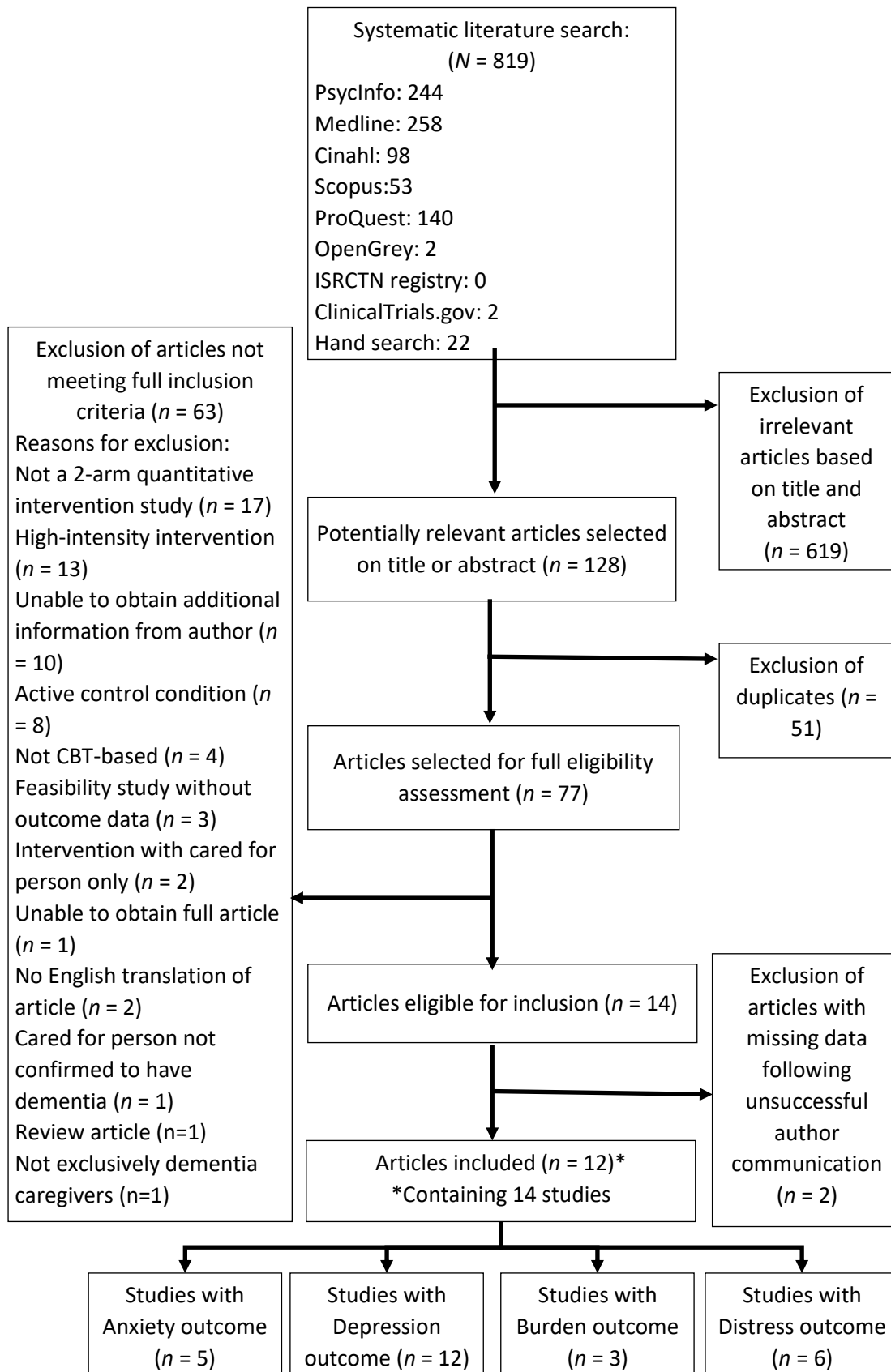


Table 1. Participant characteristics and quality of studies included in the meta-analyses

Study ID	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; NR = Not reported.

Table 2. Characteristics of interventions and outcome measures for each study

Study ID	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.

Table 3. Overview of Intervention Groups and Control Groups for Included Studies

Study ID	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.

Table 4. Quality Rating Sub-total Scores for Studies Included in the Second Empirical Paper

Study ID	Description of Participants (max = 10)	Definition and Delivery of Intervention (max = 10)	Outcome Measures			Outcome Measures Total (max = 12)	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						
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)