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Influence of positive aspects of dementia caregiving on caregivers' well-being: A systematic review

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Abstract

Background and Objectives: There is a growing evidence base that informal caregivers can identify positive aspects of providing care and that this may have a beneficial influence on their well-being. The aim of this systematic review was to explore how positive aspects of caregiving (PAC) impacts on the well-being of caregivers of people with dementia.

Research Design and Methods: We searched electronic databases for quantitative studies exploring the association between PAC and caregiver well-being. Studies were included if they involved informal (unpaid) caregivers of people with dementia, at least 75% of whom had to be residing in the community. A narrative synthesis was used to explore patterns within the data.

Results: Fifty-three studies were included in the narrative synthesis. Most studies utilized a cross-sectional design. The majority of samples consisted primarily of spouses and female caregivers. Twenty different PAC measures were employed and studies referred to a variety of constructs, such as satisfactions, gains, meaning, and rewards. PAC was associated with lower depressive symptoms and burden. Conversely, PAC was associated with better mental health, quality of life, satisfaction with life, and competence/self-efficacy. PAC was not associated with self-rated health or personal strain/stress.

Discussion and Implications: The findings suggest that identifying PAC is associated with better caregiver well-being, although further longitudinal studies are required to explore how this relationship changes over time. Interventions that enable caregivers to gain a more positive experience of caregiving could be beneficial for their well-being.

Key words: burden, gains, meaning, satisfaction, quality of life

Influence of positive aspects of dementia caregiving on caregivers’ well-being: A systematic review

Informal caregiving has been conceptualized as a career, beginning as an individual is introduced to the caregiving role and marked by transitional events (Pearlin, 1992). Many factors influence how caregivers respond to and adapt to caregiving. One way caregivers may adapt is to identify positive aspects of providing care, perceiving the potential benefits of caregiving for either themselves and/or the person cared for. A review by Kramer (1997) described how a plethora of studies had explored the ‘negative’ and ‘detrimental’ aspects of caregiving, with little attention paid to the role of positive psychological functioning in caregiving. Kramer identified that a “lack of attention to the positive dimension of caregiving seriously skews perceptions of the caregiving experience and limits our ability to enhance theory of caregiving adaption” (p.218). Whilst Kramer’s (1997b) review included all caregivers without differentiation between them based on care recipients’ health conditions, it is feasible that caregivers’ experiences may differ depending on the health condition of the care-recipient. Dementia is a progressive degenerative condition and there may be fewer opportunities for dementia caregivers to have positive experiences than caregivers of people with other health conditions (Pinquart & Sörensen, 2004). However, there is a growing evidence base concerning how identifying positive aspects of caregiving (PAC) can be beneficial for dementia caregivers’ well-being. The aim of this paper is to review the literature on positive aspects of dementia caregiving, specifically exploring the impact of PAC on caregiver well-being.

Conceptually there is no clear definition of PAC, and research indicates it may have different dimensions. For instance, caregivers have described experiencing personal growth, identifying that caregiving had made them a better person or made them more resilient (e.g., Netto, Goh, & Yap, 2009; Quinn, Clare, & Woods, 2015). Caregivers have also described feelings of gratitude and a sense of mastery (Cheng, Mak, Lau, Ng, & Lam, 2016). Within the quantitative literature, it is also clear that different terms and measures are used to describe PAC. The review by Kramer (1997b) identified the following terms used to describe PAC: satisfactions, uplifts, rewards, gratifications, growth, meaning, and enjoyment. Many of these terms lack a theoretical basis so it is difficult to determine whether they constitute separate or overlapping concepts. A range of measures of PAC have been developed using a variety of approaches. Some of these measures are based on existing tools; for instance, the Positive Aspects of Caregiving Scale (PACS; Tarlow et al., 2004) is based on the Caregiving Satisfaction Scale (CSS; Lawton, Kleban, Moss, Rovine, & Glicksman, 1989), which in turn was based on other existing measures. Other measures have been developed through identifying domains and questions from the existing literature (e.g., Faba, Villar, & Giuliani, 2017). A few have involved caregivers in the measure-development process (e.g., Abdollahpour, Nedjat, Noroozian, Salimi, & Majdzadeh, 2017) or generated questions from interviews with caregivers (e.g., Farran, Miller, Kaufman, Donner, & Fogg, 1999; Yu et al., 2016).

Theoretical models and constructs underpin understandings about PAC. Models of stress and coping have incorporated positive psychological functioning. For instance, the adapted stress and coping model (Folkman, 1997) proposed that both positive and negative emotions can co-occur in response to challenging circumstances. In this model, positive psychological states were associated with searching for and finding positive meaning, a form of coping. Finding meaning relates to the ability to identify something positive in adversity, helping the person to make sense of the situation and accept what has happened. This could result in the person being able to identify positive life changes (Park, 2010). However, in the ‘broaden-and-build’ theory, Fredrickson, Tugade, Waugh, and Larkin (2003) proposed that positive emotions have a more adaptive role in dealing with stressful circumstances, as they can provide an emotional break, replenishing depleted resources. Positive emotions can also be involved in cognitive broadening, widening people's attention, thinking, and behavior. Longitudinally this broadening effect fosters the building of a range of adaptive and durable personal resources (Fredrickson, 2004). Similarly, theories of benefit-finding imply that this emerges over time as a way of adapting to stressful circumstances. In the early stages, benefit-finding may be considered to be a form of coping, but over time may reflect positive change or growth (Tennen & Affleck, 2002).

Theoretical models of caregiving have also explored the influence of PAC on caregiver well-being. In the Stress Process Model (SPM; Pearlin, Mullan, Semple, & Skaff, 1990), gain is encompassed under 'secondary intrapsychic strains', which relates to the caregiver's self-concept. In this context, the diminishment of, or barriers to the development of feelings of, gain is considered to constitute strain. Based on two-factor models of psychological well-being, which distinguish between positive and negative aspects of psychological well-being, two-factor models of caregiving (Kramer, 1997b; Lawton, Moss, Kleban, Glicksman, & Rovine, 1991) acknowledge that caregiving can have both positive and negative outcomes. In these models, PAC is associated with positive dimensions of well-being, whilst negative aspects are associated with negative dimensions of well-being. However, Lawton et al. (1991) reported support for the two-factor model only for spousal caregivers and not for adult-child caregivers. These results highlight the inconsistent findings regarding the association between PAC and positive and negative dimensions of caregiver well-being.

A comprehensive understanding of the influence of PAC on caregiver well-being requires a systematic synthesis of the existing literature. Previous reviews have included mixed samples (Carbonneau, Caron, & Desrosiers, 2010; Kramer, 1997b) or focused on caregivers of older people (Pinquart & Sörensen, 2004). Of the reviews that have focused only on dementia caregivers, the review by Lloyd, Patterson, and Muers (2016) referred to the qualitative literature and the review by Quinn, Clare, and Woods (2010) identified the literature on finding meaning. The integrative review by Yu, Cheng, and Wang (2018) included both qualitative and quantitative studies, with a focus on the nature of PAC and factors predicting PAC. To date no review has specifically explored the association between PAC and caregiver well-being. The aim of this systematic review was to explore how PAC impacts on the well-being of caregivers of people with dementia. As part of the review process we also explored the theoretical underpinnings of the included studies and, related to this, the measures of PAC employed in the studies.

Method

The review protocol was registered with PROSPERO: CRD42017059919. The following electronic databases were searched for studies from inception to March 2017: MEDLINE (via OvidSp), PsycINFO (via OvidSp), ASSIA (via ProQuest), SSCI (via Web of Science), and CPCI (via Web of Science). Sources of grey literature were also searched, including the British Nursing Index (BNI; via ProQuest), and CINAHL (via EBSCO). An example of the search terms can be found in Supplementary document 1. Forward and backward citation searching was used to identify additional studies from relevant retrieved papers. Endnote X7 was used for reference management.

Inclusion and exclusion criteria

No date restrictions were applied, but studies had to be published in English. The inclusion criterion for caregivers was that they had to be informal (unpaid) caregivers of people with dementia, and we excluded studies where over 25% of the people with dementia had died. The person with dementia could have any dementia diagnosis, though studies of people with Mild Cognitive Impairment were excluded. Studies with mixed samples were included if the data for the person with dementia were provided separately: if the data were not presented separately, at least 75% of the sample had to have a diagnosis of dementia. At least 75% of the people with dementia had to be community-dwelling (calculated at baseline in longitudinal studies). In many papers, the place of residence of the person with dementia was not stated or was unclear and in these circumstances it was inferred that they were community dwelling if this was implied by the other data reported. For instance, some papers noted that caregivers were providing over four hours of care a day, which suggests they were residing with the person with dementia.

Studies reporting cross-sectional or longitudinal associations between PAC and caregiver well-being were included. PAC was defined as the caregiver deriving something positive out of providing care; thus, positive aspects had to be directly related to caregiving. Caregiver well-being is a multi-dimensional concept (Manthorpe & Bowling, 2016) and global measures, such as quality of life (QoL) and satisfaction with life, as well as caregiver-specific well-being measures, such as stress, burden, role strain, and competence, were included.

Review process

Figure 1 illustrates the literature search process. Title, abstract, and full-text screening were conducted by two reviewers. Where there was uncertainty on a study it was included in the next stage of screening. There was 86~~.2~~% agreement on title screening, 84~~.2~~% agreement on abstract screening, and 82% agreement on full-text screening. Any disagreements were resolved through discussion. Study information was extracted from the included papers by the principal reviewer using a structured proforma and checked by the second reviewer. Details of the information extracted from the papers are provided in Supplementary document 2.

Study quality

Study quality was assessed using the QATSDD (Sirriyeh, Lawton, Gardner, & Armitage, 2012) and we used the 14 indicators in the tool that applied to quantitative studies (two items are applicable only to qualitative studies). Indicators were rated on a 4-point Likert scale, with possible total scores ranging from 0 to 42; higher scores indicated higher quality reporting. Study quality was rated by two reviewers and discussed until consensus was reached. In six instances, data from a study were reported in two papers. If data were reported in a thesis/dissertation and a published paper, only the published paper was included in the review. When data were reported in two published papers, the higher scoring (or most detailed) paper was included in the analysis. One paper obtained a particularly low score (Uwakwe, 2006), but was not excluded as it was published as a short research letter and the word limit may have constrained the amount of information provided.

Narrative synthesis

Due to the heterogeneity of study designs and outcome measures, a narrative synthesis was used. A narrative synthesis approach involves the synthesizing of findings using a textual approach to discuss the findings of the review. In this study it was used to review the nature and direction of effects and explore patterns within the data. A similar approach to that reported in Farina et al. (2017) was followed. In the narrative synthesis, findings in the included studies were explored to see whether they reported a significant association with the outcome measures and if so in what direction. Non-significant associations were also recorded. The findings explored were the results from correlation, regression, or other analytical techniques that explored associations between variables. When studies reported regression analyses, it was noted whether PAC was a predictor of the outcome measure.

Results

Fifty-nine papers reporting 53 studies were included in the narrative synthesis (reported in Supplementary document 3). Papers were published between 1989 and 2017 and the majority employed cross-sectional designs: only five papers used longitudinal designs. Most studies were conducted within America and Canada (reported in Table 1).

Paper quality

No paper achieved the maximum score on the QATSDD (reported in Table 2). The overall range of ratings was 11-38, and 49% of papers (N = 26) achieved ratings of between 31 and 35. All papers achieved maximum scores for selecting an appropriate data collection method and 98% (N = 52) achieved maximum scores for selecting an appropriate data analysis method. The majority of studies (83% [N = 44]) provided clear evidence that they recruited representative samples (i.e., they achieved a maximum score of 3 for this criterion). There was significant variation across studies in the extent to which the reliability and validity of the measures had been considered (only 30% [N = 16] of studies achieved maximum scores whilst 23% [N = 12] scored between 0-1 on this criterion). Similarly, there was variation in the reporting of data collection procedures (32% [N = 17] of studies achieved maximum scores whilst 9% [N = 5] scored between 0-1 on this criterion). However, no paper provided evidence of patient/public involvement and 55% (N = 29) did not provide any justification for the sample size.

Participants

The majority of samples (n = 51) comprised over 50% female caregivers. Most caregivers were aged over 65 years. The majority of caregivers were spouses of the person with dementia (represented in 83% [n = 119] of associations), followed by adult child or daughter/ son in-laws (represented in 76% [n = 109] of associations). The duration of caregiving was difficult to extrapolate: several papers did not contain this information. Where stated, the average duration of caregiving ranged from 2.6 to 5.6 years.

Details about the person with dementia were often not reported. The most commonly specified diagnosis was Alzheimer's disease (represented in 57% [n = 81] of associations). Only 20 papers contained information about the severity of the person's dementia: the majority fell in the moderate (41% [n = 11]) and moderately severe (30% [n = 8]) stages with fewer being in the early/ mild stage (15% [n = 4]) or severe stage (15% [n = 4]). Most studies recruited through support services including health services or charities such as the Alzheimer's Association. Only a minority of studies recruited participants directly from the community, such as through churches or community centers.

Theoretical basis

The majority of papers (83% [n = 44]) made reference to theory, models, frameworks, and constructs. Some papers referenced multiple models, constructs, and theories; thus, the total number referenced was 76. However, only 60% of papers (n = 32) provided a specific theoretical basis for the study reported or applied a theoretical framework, model, or construct to the research findings. Furthermore, nine studies made no reference to any theories, models, or constructs.

The majority of models referenced (36% [n = 27]) related to the positive and negative aspects of caregiving with the dominant model being the stress and coping model (Lazarus & Folkman, 1984) and its revision (Folkman, 1997), referenced on 12 occasions. The SPM and its revision (Pearlin et al., 1990) was referenced on eight occasions and the two-factor model of caregiving (Lawton et al., 1991) was referenced on four occasions. Generic models and theories of stress and adaptation were frequently referenced (n = 13) but only the sociocultural stress and coping model (Knight, Silverstein, McCallum, & Fox, 2000) was referenced by more than one paper (n = 2). Some models (n = 8) concerned the relationship dynamics and interactions between caregivers and the care recipient or environment. For instance, the ABCX family crisis model (McCubbin & Patterson, 1983), which explores family's adjustment and adaptation to stressful events, was referenced on three occasions. However, only one paper referenced the broaden-and-build theory of positive emotions (Fredrickson, 2004).

Measures of PAC

The titles of the measures employed in the studies infer that various constructs were captured, including finding meaning, satisfaction, gains, uplifts, rewards, esteem, gratification, and coping. A total of 20 different of PAC measures were used in the studies, and these ranged in length from four items to 110 items, the majority having ten items or fewer. The Positive Aspects of Caregiving Scale (PACS; Boerner, Schulz, & Horowitz, 2004; Tarlow et al., 2004) was most commonly used, being included in 18 studies (and in 29% [n = 40] of associations). The Caregiving Satisfaction Scale (CSS; Lawton et al., 1989) and its revised version were used in eight studies (and in 15% [n = 21] of associations). The Finding Meaning Through Caregiving Scale (FMTCS; Farran et al., 1999) was used in four studies (and in 11% [n = 15] of associations) and Strawbridge's caregiving satisfaction scale (Strawbridge, 1991), and its adapted version were used in four studies (and in 6% [n = 8] of associations). Even when the same measure was used by studies, variation in the items used were apparent. For instance, the original PACS has 11-items, the revised scale has nine items and there is a further Chinese version. Sixteen other measures were also used, and four of these were study-developed. Sometimes two measures of PAC were used and this occurred in five studies. In three instances the purpose of this was to validate a newly-developed PAC measure. However, two other cross-sectional studies included measures of both satisfaction and gain. Finally, three linked studies employed a qualitative approach, which asked caregivers to report the most enjoyable aspects of their role and then quantified these responses for the purposes of analysis.

Associations between positive aspects of caregiving and caregiver well-being

A total of 143 relevant associations were extracted from the 53 studies. Most papers used standardized measures (i.e., measures with specified guidance to enable consistent and comparable administration and scoring); the only anomaly was that in 75% (n = 12) of the associations concerning health, a self-report item was used. Half of the outcome measures used (n = 19) were caregiver-specific. The most frequent association explored was with burden (28% [n = 40]). Other commonly explored associations were with depression (18% [n = 26]) and mental health/ psychological health 18% [n = 25]). Findings for each well-being category are presented below and summarized in Supplementary Table 1.

Global measures of wellbeing.

Depressive symptoms. Twenty-six associations explored the relationship between PAC and depression symptoms and 81% of these were significant. The weight of evidence suggests that higher PAC is associated with reporting fewer symptoms of depression. This association was explored in a wide range of caregiving relationships. In most cases, where this information was reported, the person with dementia was diagnosed with Alzheimer’s disease. All these associations employed cross-sectional data and the most frequently used measure was the PACS (n = 11), but a total of seven different measures were employed.

Mental health. Twenty-five associations explored the relationship between PAC and mental health and 80% of these were significant. A connection between PAC and better mental health is suggested. Positive associations were reported for psychological wellbeing, psychological health, mental health, and positive affect. In comparison, negative associations were found for mental health problems, anxiety, psychological distress, depression, negative emotional reactions, and negative affect. These associations were explored in a range of caregiving relationships and dementia diagnoses. Sixteen studies used a cross-sectional design and nine used longitudinal data. The most common means of capturing PAC was to quantify caregiver qualitative reports. However, eight other standardized measures were also used.

QoL/ Health-related QoL. Nine associations explored the relationship between PAC and QoL/health-related QoL and 89% of these achieved significance. The majority of the evidence suggests that reporting higher PAC is associated with higher QoL. The one significant association with poorer QoL came from a European multi-site study by Alvira et al. (2015), which reported associations between positive aspects of caregiving and QoL from each study site. The significant negative association was found in Estonian data but this represents only 6% of the QoL associations reported in this study. Associations with QoL were explored in a range of caregiving relationships. However, all these associations came from cross-sectional studies and, when stated, most people with dementia were diagnosed with Alzheimer’s disease. Five different measures of PAC were used, with the PACS (n = 4) employed most frequently.

Satisfaction with life. Four associations explored the relationship between PAC and satisfaction with life and 75% of these achieved significance. Reporting higher PAC is predominately associated with greater satisfaction. This association was explored in a range of caregiving relationships using cross-sectional designs. When the information was provided, all people with dementia were diagnosed with Alzheimer's disease and four different PAC measures were employed.

Health. Sixteen associations explored the relationship with PAC and health and 38% of these were significant. The balance of evidence suggests that there is no significant association between PAC and self-reported health. This association was explored using cross-sectional designs in a range of caregiving relationships and types of dementia. Eight different measures of PAC were used, with the Strawbridge caregiver satisfaction scale (n = 4) employed most.

Caregiver-specific wellbeing measures

Burden. Forty associations explored the relationship between PAC and burden and 85% of these were significant. The majority of studies suggest that higher PAC is associated with less burden. This association was explored in a wide range of caregiving relationships. People with dementia had a range of diagnoses, including some of the rarer forms of dementia. Longitudinal as well as cross-sectional data were interrogated. Fifteen different measures of PAC were used to explore this association, with the PACS being the most commonly used (n = 12).

Role strain. This category includes constructs related to role overload, role captivity, and negative feelings about the caregiving role. Eleven associations explored the relationship between PAC and role strain and 46% % of these were significant. The balance of evidence suggests that no conclusions can be drawn on the association between PAC and role strain. This association was explored in a range of caregiving relationships and different dementia diagnoses. Only one study employed a longitudinal design, with the rest being cross-sectional. Of the seven different PAC measures used, the most common was the PACS (n = 3).

Personal strain/ Stress. Five associations explored the relationship between PAC and stress and 40% of these were significant. The balance of evidence suggests no significant association between PAC and stress or personal strain. This association was explored in a range of caregiving relationships and dementia diagnoses. Five different measures of PAC were used but associations have been based on cross-sectional data only.

Competence/ Self-efficacy. Seven associations explored the relationship between PAC and competence and 71% of these were significant. There is some evidence that reporting higher PAC is associated with higher competence or self-efficacy. This association was explored in a range of caregiving relationships using cross-sectional data. Out of the five measures of PAC, the most commonly used were the PACS (n = 2) and the Meaning in caregiving scale (n = 2).

Discussion

This is the first review to explore the impact of PAC on dementia caregiver well-being comprehensively. Overall the findings indicate that being able to identify PAC is associated with higher caregiver well-being. The available evidence indicates that PAC was associated with lower depressive symptoms and burden. It was also associated with better mental health, QoL, satisfaction with life, and competence/self-efficacy. The balance of evidence indicates that PAC is not associated with caregiver self-rated health. However, self-rated health is likely to be impacted by many factors and caregivers’ appraisal of PAC may have less influence than other determinants such as caregiver health condition or daily functioning. Despite PAC being associated with burden, PAC was not significantly associated with personal strain/ stress and there was inconclusive evidence about the association with role strain. This finding may be a reflection that more studies have explored the association with burden or it could suggest that personal strain/ stress, and role strain are conceptually different from caregiving burden. There were some inconsistencies in the findings; for example, not all studies reported a significant association between PAC and depression. One explanation for these inconsistencies is that the studies varied in the measures of PAC employed and also in the outcome measures used. Thus, the measures employed may have had an influence on the results. In addition, studies varied in the sample sizes; studies with smaller samples may not have had enough power to detect a statistically significant result.

The majority of studies referred to theories, frameworks, models, or constructs. However, only 60% of papers used these concepts as a basis for the research study. Various models were referenced, with most authors referencing stress-coping frameworks. The SPM (Pearlin et al., 1990) was the most commonly cited caregiving model. However, the SPM does not provide a clear role for PAC. The two-factor model, referenced in four papers, conceptualizes PAC as a form of appraisal linked to positive dimensions of well-being. Yet, the findings of this review indicate that PAC was associated with both positive and negative dimensions of well-being. Other papers referenced more generic theories of stress and coping (Folkman, 1997; Lazarus & Folkman, 1984), in which PAC is also viewed as a form of appraisal or a method to mitigate the effects of caregiving stress. Thus, in these models, PAC is viewed more as a moderator of the caregiving experience. Furthermore, some studies referenced models, in which PAC is perceived as an outcome of caregiving, such as the ABCX family crisis model which concerns families' adaptability to stressful circumstances. In the ABCX model, PAC can be a positive outcome, or 'bonadaptation' (Kramer, 1993). These findings suggest that there is a need for further caregiving models to be developed that fully encompass the role of PAC. This may involve building on positive psychological approaches:

only one included paper (DeGregory, 2014) drew on the broaden-and-build theory of positive emotions.

The findings of this review are consistent with Kramer’s (1997b) conclusion that there is a lack of conceptual clarity around the definition of PAC. Examination of the measures employed in the studies indicates the main domains being investigated were: satisfactions, gains, meaning, and rewards. It is difficult to determine whether these measures are tapping into different dimensions of PAC or similar constructs, particularly as not all measures are published. Further, the names of measures may not necessarily reflect their content. For instance, it is possible that the GAIN and BENEFIT measures (Lawton et al., 1991; Lawton, Rajagopal, Brody, & Kleban, 1992) were the same measure, as they both contained the same number of items. Interestingly, in two papers (Morano, 2003a, 2003b) measures of both gain and caregiving satisfaction were included, implying these were considered to be separate constructs. The most commonly used measure was the PACS. However, the popularity of this measure may be due to it being included in the large-scale multi-site REACH and REACH II intervention studies; many of the included studies used these datasets for analyses. The review identified that, in recent years, new measures of PAC have been were developed (Faba et al., 2017; Yu et al., 2016), including more culturally-specific measures (Abdollahpour et al., 2017).

The majority of studies employed cross-sectional designs; thus, there is little longitudinal information about the association between PAC and well-being. The majority of studies used samples of convenience; caregivers were often recruited through support services, health services, or charities with only a few studies directly recruiting from the community. Approaching caregivers directly in the community may have enabled researchers to access people who were not accessing any formal support. It is possible these caregivers do not feel the need to access support services because they feel more positive about their role, or conversely they may be more negative about their role because they are not in contact with any external formal support. In terms of caregiver characteristics, many were in the early stages of their caregiving career. Unfortunately, the samples were often heterogeneous, which might hide meaningful differences. For instance, although most participants were spousal caregivers, it was not unusual for studies to include up to seven different forms of caregiving relationship within the sample. Probably the experience of PAC differs depending on the type of caregiving relationship (e.g., Broese van Groenou, de Boer, & Iedema, 2013). Furthermore, most participants were women: only Baker, Robertson, and Connelly (2010) and Kramer (1997a) focused solely on male caregivers. The lack of male caregivers hinders the exploration of meaningful gender comparisons.

In considering the findings, it is important to recognize the review's strengths and limitations. At the abstract screening stage, we identified four dissertations of which full-text versions could not be located. However, published papers from two of these dissertations were included in the review. The focus of this review was on the association between PAC and caregiver well-being; thus, factors predicting PAC were not explored. As two recent systematic reviews have explored the qualitative literature (Lloyd et al., 2016; Yu et al., 2018) this review focused on the quantitative literature. As in other reviews (e.g., Farina et al., 2017) a narrative synthesis approach was taken, given the heterogeneity in the included studies. Although this review included both published papers and unpublished dissertations/theses, there is a risk of publication bias, as it is possible that studies reporting significant associations between PAC and other measures are more likely to be published. Furthermore, studies often included multiple measures, but some did not report the associations between all of these measures. Eight of the included studies used data from the REACH and REACH II studies, so the same dataset may have been used in multiple studies.

In comparison to the number of tools available for rating the quality of randomized controlled trials and qualitative studies, there seems to be a dearth of tools for rating cross-sectional quantitative studies. The QATSDD (Sirriyeh et al., 2012) was selected because the items seemed appropriate for the types of papers included; however, there have been criticisms that the QATSDD is too subjective (Fenton, Lauckner, & Gilbert, 2015). Whilst there were clearly benefits in using the QATSDD, there were also challenges to implementing the tool. For instance, we found that studies with smaller word counts (because of journal requirements) risked having a lower score because there is less scope to explain the study in-depth. This suggests that quality rating tools would benefit from more flexibility: for instance, the ability to take into account the length of the paper.

The findings of the review identify areas which require further investigation. First, we used a broad definition of well-being, which included both caregiver-specific measures and more global measures. The majority of the well-being measures used focused on the 'negative' aspects of well-being, such as burden and depression. This reflects the wider dementia caregiving literature where the majority of research has focused on specific domains of well-being (Manthorpe & Bowling, 2016). Few studies explored the association between PAC and more 'positive' global measures of QoL, satisfaction with life, or well-being. Thus, there needs to be more research exploring these associations, particularly as QoL is a commonly used outcome in intervention research (Pendergrass, Becker, Hautzinger, & Pfeiffer, 2015). Additionally, few studies have explored PAC longitudinally, and research is needed to explore whether experiences of PAC change throughout the caregiving career and, if so, how this influences well-being. The broaden-and-build theory states that over time positive emotions broaden a person’s way of thinking. This implies that the adaptive effects of positive emotion occurs longitudinally and may play a greater role later on in the caregiving career.

The findings of this review suggest that being able to identify PAC seems to be beneficial for caregiver well-being. Healthcare professionals providing support for caregivers need to consider and recognize that caregivers can have both positive and negative experiences. The findings also raise the possibility that PAC can be targeted through interventions. Whilst interventions may not be able to directly enable caregivers to experience PAC, caregivers might be helped to appraise their situations more positively, resulting in a better experience of caregiving. Cheng et al. (2012) developed the ‘Benefit-finding intervention’ program, which promoted the use of positive appraisal to engender benefit-finding. Building on a psychoeducational program, caregivers in the intervention completed exercises on positive re-appraisal; identifying stressful situations and then re-evaluating them to provide more positive appraisals. Compared to the control groups (receiving psycho-educational programs), caregivers in the intervention reported lower depression symptoms post-intervention (Cheng et al., 2017). Similarly, a multi-media support intervention, which also targeted appraisals of caregiving, was effective in increasing reports of PAC and caregiving competence (Beauchamp, Irvine, Seeley, & Johnson, 2005). Interventions may also be able to indirectly improve experience of PAC through targeting the caregiving situation. For example, Savundranayagam (2014) found that increases in the amount of help received from others, as well as satisfaction with this help, were both associated with an increase in PAC in the form of positive attitudes towards the dementia caregiving role.

Conclusion

The review findings suggest that identifying PAC is associated with better caregiver well-being. There is an increasing evidence base for the role of PAC in the dementia caregiving experience, although gaps in the literature should be addressed. A more consistent terminology and approach to the conceptualization and measurement of PAC is needed. The majority of papers relied on stress-coping frameworks and there is a need for caregiving models to be developed that fully encompass the role of PAC. This may involve building on concepts from positive psychology. There was heterogeneity in the samples and most studies relied on cross-sectional designs; further longitudinal studies are required to explore how experiences of PAC change over time. The findings have important implications for the development of interventions and supportive services for caregivers. Interventions that help caregivers gain a more positive experience of caregiving could be beneficial for their well-being.

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Table 1. Geographical regions represented in publications

Geographical region	N of publications
America and Canada	32
New Zealand	1
Europe (including the United Kingdom)	8
East Asia	10
Middle East	1
Africa	1

Table 2. Summary of studies included in the review

Reference	Design	Sample size	Positive aspects of caregiving measures	Wellbeing measures[1]	Quality rating
Abdollahpour et al., 2017	Measure validation	132	Positive aspects of caregiving questionnaire- 10 items	SR health Iranian caregiving burden questionnaire	31
Alvira et al., 2015	Cross-sectional	2014	Caregiver esteem subscale- 7 items	ZBI EQ-5D GHQ	28
Andren & Elmstahl, 2005	Cross-sectional	153	CASI	Nottingham health profile scale CBS	29
Baker et al., 2010	Cross-sectional	70	Strawbridge caregiving satisfaction scale-5 items	SR health ZBI	25
Cheng et al., 2013	Cross-sectional	99	PACS	ZBI Role overload measure Hamilton depression rating scale	35
Cohen et al., 1994	Longitudinal measure validation	196 baseline	Asked caregivers about most enjoyable aspects of role, which were then quantified	GHQ ZBI	24
de Labra et al., 2015	Cross-sectional	101	Revised CSS	ZBI Caregiving competence scale	25
DeGregory, 2014	Cross-sectional	55	PACS	SwLS WHOQOL-BREF	36
Faba et al., 2017	Measure validation	260 (study 2)	GAC	SwLS GDS-SF ZBI	30

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[1] Only the well-being measures that were used in the analyses with the PAC measures were extracted.

Reference	Design	Sample size	Positive aspects of caregiving measures	Wellbeing measures[1]	Quality rating
Farran et al., 1999	Measure validation	215 (study 2)	Provisional meaning subscale- 19 items, FMTCS	CES-D Global role strain scale Personal gain measure Caregiver satisfaction measure	24
Gold et al., 1995	Longitudinal	196 baseline	Asked caregivers about most enjoyable aspects of role, which were then quantified	GHQ ZBI	28
Goncalves-Pereira et al., 2010	Cross-sectional	116	PACS	ZBI GHQ	31
Harris et al., 2011	Cross-sectional	621	PACS	CES-D ZBI	33
Harwood et al., 2000	Cross-sectional	64	CSS- 5 items	SF-36 CES-D CBS	24
Heo, 2014	Cross-sectional	648	PACS- 9 items	CES-D ZBI	35
Hilgeman et al., 2007	Cross-sectional	243	PACS- 9 items	CES-D	34
Kajiwara et al., 2015	Cross-sectional	354	Caregiving gratification scale- 8 items	ZBI- Japanese version	30
Kinney & Stephens, 1989	Cross-sectional	60	Caregiving hassles and uplifts scale- 110 items appraised as hassle or uplift	SCL-R-90-R	21
Kramer, 1993	Cross-sectional	72	Strawbridge caregiving satisfaction scale- 15 items	SR health	33
Kramer, 1997	Cross-sectional	74	Strawbridge caregiving satisfaction scale- 15 items	SR health Screen for caregiver burden	32

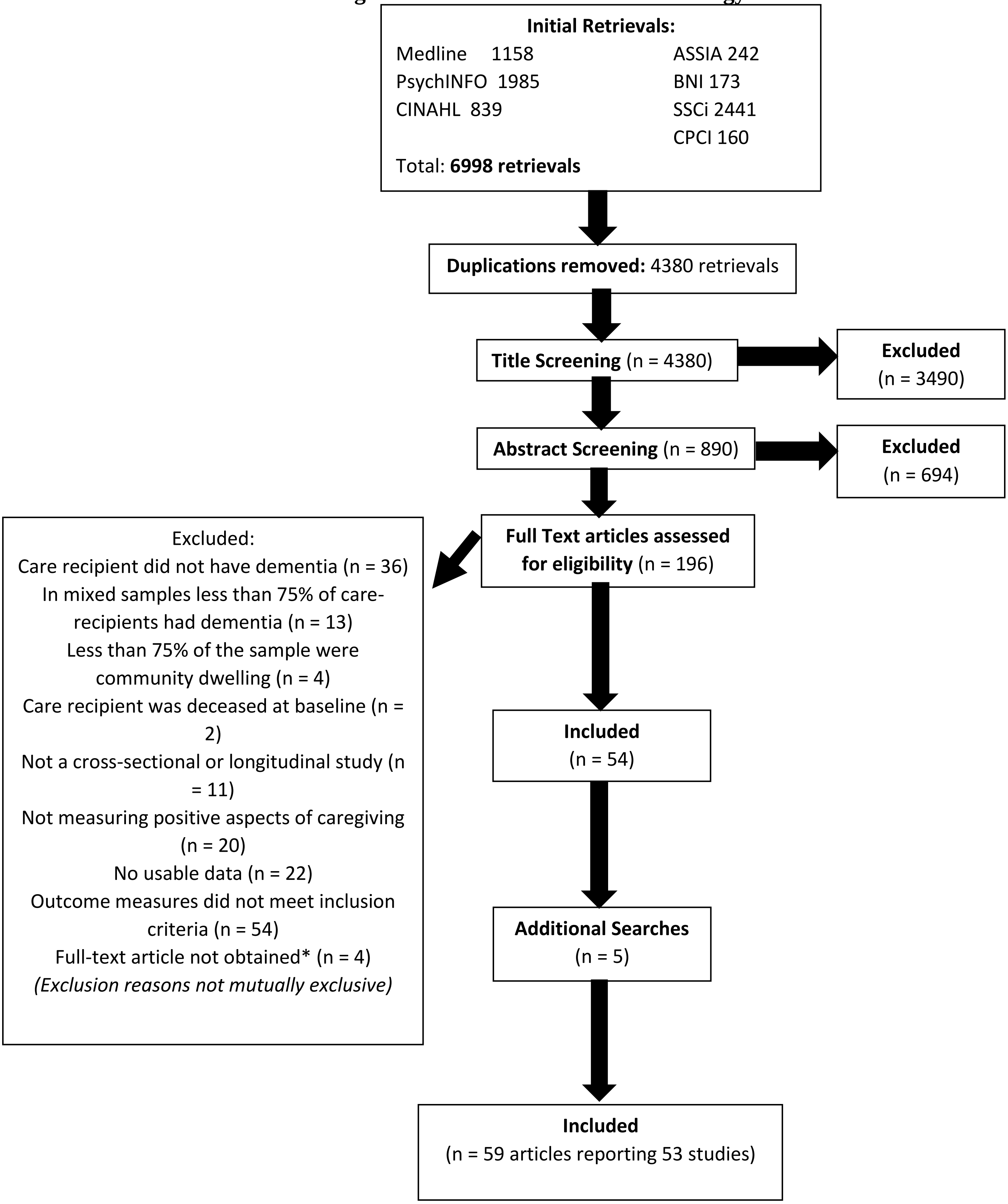
Reference	Design	Sample size	Positive aspects of caregiving measures	Wellbeing measures[1]	Quality rating
Lawton et al., 1991	Cross-sectional	632	CSS- 5 items	SR health Subjective caregiving burden Affect balance scale CES-D	30
Lawton et al., 1992	Cross-sectional	629	CSS- 5 items	SR health Subjective caregiving burden Affect balance scale	32
Lethin et al., 2017	Longitudinal cohort	1223 in total	Caregiver esteem subscale- 7 items	GHQ	25
Lévesque et al., 1995	Cross-sectional	265	Satisfaction with caregiving role- 5 items	Brief symptom inventory Negative feelings about caregiving role scale Affect balance scale	32
Lévesque et al., 1998	Longitudinal	265 baseline	Satisfaction with caregiving role- 5 items	Brief symptom inventory Negative feelings about caregiving role scale Affect balance scale	32
Liew et al., 2010	Cross-sectional	334	GAIN- 10 items	GHQ ZBI SSCQ	30
Liu, 2009	Cross-sectional	257	PACS- 9 items	CES-D	31
Liu et al., 2012	Cross-sectional	96	PACS-9 items	ZBI SF-36	35
Lloyd, 2008	Cross-sectional	64	Provisional meaning subscale- 19 items, FMTCS	CES-D	34
Lou et al., 2015	Measure validation	374	PACS- 11 items, Chinese version	CES-D SR health ZBI	32

Reference	Design	Sample size	Positive aspects of caregiving measures	Wellbeing measures[1]	Quality rating
Mbiza, 2016	Cross-sectional	643	PACS- 11 items	CES-D SR health	38
McLennon et al., 2011	Cross-sectional	84	FMTCS- 43 items	SF-36 ZBI	38
Monin et al., 2015	Cross-sectional	58	PACS- 11 items	ZBI CES-D	32
Morano, 2003	Cross-sectional	204	CSS- 5 items Personal gain- 4 items	CES-D Life satisfaction	31
Morano, 2003b	Cross-sectional	103	CSS- 5 items, Personal gain- 4 items	CES-D Life satisfaction	34
Narayan et al., 2001	Cross-sectional	50	PACS- 11 items	Caregiving competence scale Role captivity scale	32
Picot, 1991	Cross-sectional	83	Caregiver rewards scale- 24 items	Cost of care index	37
Plata, 2007	Cross-sectional	60	FMTCS- 43 items	SR health ZBI CES-D GHQ Quality of life	36
Quinn et al., 2012	Cross-sectional	447	Meaning in caregiving scale- 12 items	Role captivity scale Caregiving competence scale SR health	33
Rapp & Chao, 2000	Cross-sectional	63 (in data analysis)	Appraisals of gain- 11 items	SR health ZBI Positive and negative affect	26
Rapp et al., 1998	Cross-sectional	65	BENEFIT- 11 items	SR health Quality of life CES-D	28

Reference	Design	Sample size	Positive aspects of caregiving measures	Wellbeing measures[1]	Quality rating
Reis et al., 1994	Longitudinal	213 baseline	Asked caregivers about most enjoyable aspects of role, which were then quantified	ZBI GHQ	27
Roff et al., 2004	Cross-sectional	618	PACS- 9 items	CES-D Speilberger state trait personality inventory	33
Roud et al., 2006	Measure validation	45	Personal gain scale- 6 items Positive value, 5 items, COPE index	GHQ Burden interview Caregiving competence scale	22
Semiatin & O'Connor, 2012	Cross-sectional	57	PACS- 9 items	CES-D RIS Eldercare self-efficacy scale	29
Shyu et al., 2010	Cross-sectional	176	Rewards of caregiving scale- 14 items	FCI role strain scale SF-36 CES-D Chinese version	35
Son et al., 2003	Cross-sectional	117	Strawbridge caregiving satisfaction scale- 16 items	SR health Korean burden inventory	34
Talkington-Boyer & Snyder, 1994	Cross-sectional	110	CSS- 5 items	Subjective caregiver burden scale Schwab, Holzer & Warheit (1973) depression scale	20
Uwakwe, 2006	Cross-sectional	30	CSS- 5 items	RSS ZBI	11
Williams, 2005	Cross-sectional	720	PACS- 11 items	CES-D	32
Yap et al., 2010	Measure validation	238	GAIN- 10 items PACS- 9 items	ZBI	30
Yu et al., 2016	Cross-sectional	401	PACS- 9 items, Chinese version	ZBI	33
Yu et al., 2015	Cross-sectional	168	PACS- 9 items	Caregiving burden interview- Chinese version	31

Note. BENEFIT- Perceived Role Benefit Score; CASI- Carers Assessment of Satisfaction Index; CBS- Caregiving Burden Scale; CES-D- Centre for Epidemiologic Studies- Depression scale; CSS- Caregiving Satisfaction Scale; EQ-5D- EuroQol-5D; FMTCS- Finding Meaning Through Caregiving Scale; GAC- Gains Associated with Caregiving; GAIN- Gains in Alzheimer's Care Instrument; GDS-SF- Geriatric Depression Scale- Short Form; GHQ- General Health Questionnaire; PACS- Positive Aspects of Caregiving Scale; RSS- Relatives Stress Scale; SF-36- Short Form- 36; SR- Self Reported; SSCQ- Short Sense of Competence Questionnaire; SwLS- Satisfaction with Life Scale; WHOQOL-BREF- World Health Organization Quality of Life assessment- Bref; ZBI- Zarit Burden Inventory;

Figure 1. Flowchart of the search strategy



Supplementary document 1: Example search strategy for OVID

1	dementia/ or aids dementia complex/ or dementia with lewy bodies/ or presenile dementia/ or semantic dementia/ or senile dementia/ or vascular dementia/ or alzheimer's disease/ or cognitive impairment/ or corticobasal degeneration/ or creutzfeldt jakob syndrome/ or melas/ or neurodegenerative diseases/ or neurofibrillary tangles/ or parkinson's disease/ or picks disease/ or pseudodementia/ or senile plaques/
2.	exp caregivers/
3.	"positive aspects of care"
4.	"positive appraisal"
5.	"Positive experience"
6.	"Positive perception"
7.	"Positive impact"
8.	"positive outcome"
9.	enjoyment*
10.	satisfaction*
11.	meaning*
12.	benefit*
13.	gratification
14.	pleasure or gain*
15.	uplift*
16.	strength
17.	reward
18.	"personal growth"
19.	"positive aspects of care".mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
20.	3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19
21.	1 and 2 and 20
22.	limit 21 to English language

Supplementary document 2: Details of information extracted from the papers

1. Type of study
2. Theoretical/conceptual model/constructs
3. Setting e.g. community
4. Geographical location of study
5. Number of caregivers
6. Gender of caregivers
7. Age of caregiver
8. Caregiver relationship to person with dementia
9. Length of caregiving
10. Gender of person with dementia
11. Age of person with dementia
12. Diagnosis
13. Dementia severity
14. Measure of positive aspects of caregiving
15. Well-being measures
16. Salient results

Supplementary document 3: Publications included in the systematic review

1. Abdollahpour, I., Nedjat, S., Noroozian, M., Salimi, Y., & Majdzadeh, R. (2017). Positive Aspects of Caregiving Questionnaire: A validation study in caregivers of patients with dementia. *Journal of Geriatric Psychiatry and Neurology, 30*(2), 77-83. doi:10.1177/0891988716686831
2. Alvira, M. C., Risco, E., Cabrera, E., Farre, M., Rahm Hallberg, I., Bleijlevens, M. H., . . . RightTimePlaceCare, C. (2015). The association between positive-negative reactions of informal caregivers of people with dementia and health outcomes in eight European countries: A cross-sectional study. *Journal of Advanced Nursing, 71*(6), 1417-1434. doi:10.1111/jan.12528
3. Andren, S., & Elmstahl, S. (2005). Family caregivers' subjective experiences of satisfaction in dementia care: Aspects of burden, subjective health and sense of coherence. *Scandinavian Journal of Caring Sciences, 19*(2), 157-168. doi:10.1111/j.1471-6712.2005.00328.x
4. Baker, K. L., Robertson, N., & Connelly, D. (2010). Men caring for wives or partners with dementia: Masculinity, strain and gain. *Aging & Mental Health, 14*(3), 319-327. doi:10.1080/13607860903228788
5. Cheng, S. T., Lam, L. C., Kwok, T., Ng, N. S., & Fung, A. W. (2013). Self-efficacy is associated with less burden and more gains from behavioral problems of Alzheimer's disease in Hong Kong Chinese caregivers. *Gerontologist, 53*(1), 71-80. doi:10.1093/geront/gns062
6. Cohen, C. A., Gold, D. P., Shlman, K. I., & Zucchero, C. A. (1994). Positive aspects in caregiving - an overlooked variable in research. *Canadian Journal on Aging-Revue Canadienne Du Vieillissement, 13*(3), 378-391. doi:10.1017/s071498080000619x
7. de Labra, C., Millan-Calenti, J. C., Bujan, A., Nunez-Naveira, L., Jensen, A. M., Peersen, M. C., . . . Maseda, A. (2015). Predictors of caregiving satisfaction in informal caregivers of people with dementia. *Archives of Gerontology and Geriatrics, 60*(3), 380-388. doi:10.1016/j.archger.2015.03.002
8. DeGregory, C. (2014). *The effects of multiple gratitude interventions among informal caregivers of persons with dementia and Alzheimer's disease.* (Ph.D.), University of South Carolina.
9. Faba, J., Villar, F., & Giuliani, M. F. (2017). Development of a measure to evaluate gains among Spanish dementia caregivers: The gains associated with caregiving (GAC) scale. *Archives of Gerontology and Geriatrics, 68*, 76-83. doi:10.1016/j.archger.2016.09.004
10. Farran, C. J., Miller, B. H., Kaufman, J. E., Donner, E., & Fogg, L. (1999). Finding meaning through caregiving: Development of an instrument for family caregivers of persons with Alzheimer's disease. *Journal of Clinical Psychology, 55*(9), 1107-1125. doi:10.1002/(sici)1097-4679(199909)55:9<1107::aid-jclp8>3.0.co;2-v
11. Gold, D. P., Cohen, C., Shulman, K., Zucchero, C., Andres, D., & Etezadi, J. (1995). Caregiving and dementia: Predicting negative and positive outcomes for caregivers. *International Journal of Aging & Human Development, 41*(3), 183-201. doi:10.2190/RGYJ-5KA2-7THX-7BQ5
12. Goncalves-Pereira, M., Carmo, I., da Silva, J. A., Papoila, A. L., Mateos, R., & Zarit, S. H. (2010). Caregiving experiences and knowledge about dementia in Portuguese clinical outpatient settings. *International Psychogeriatrics, 22*(2), 270-280. doi:10.1017/s1041610209991050
13. Harris, G. M., Durkin, D. W., Allen, R. S., DeCoster, J., & Burgio, L. D. (2011). Exemplary care as a mediator of the effects of caregiver subjective appraisal and emotional outcomes. *Gerontologist, 51*(3), 332-342. doi:10.1093/geront/gnr003
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15. Heo, G. J. (2014). Religious coping, positive aspects of caregiving, and social support among Alzheimer's disease caregivers. *Clinical Gerontologist, 37*(4), 368-385. doi:10.1080/07317115.2014.907588
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Supplementary Table 1. Summary of findings from the studies identified in the review

Caregiver well-being outcomes (N associations)	Findings	Number of studies/Details	Kin-relationship	Dementia diagnosis (N)	Positive aspects of caregiving measures (N)	Design employed (N)	Quality rating *M* (range)
Depressive symptoms (26)	Associated with fewer depressive symptoms	14	Spouse Partner Child Child-in-law Sibling Grandchild Niece Relative Friend Other	AD (18) Mixed (1)	PACS (11) CSS (5) Caregiving uplifts scale (4) FMTCS (3) BENEFIT (1) Rewards of caregiving scale (1) Gains associated with caregiving (1)	Cross-sectional (23) Measure validation (3)	29.7 (20-38)
	Associated with higher depressive symptoms	2					
	No significant association	4					
	Related findings	6 Predicted variance (except in one instance).					
Burden (40)	Associated with lower burden	26	Spouse Partner Child Child-in-law Sibling Grandchild Niece Cousin Relative Friend Other	AD (22) Mixed (3) Vascular (10) DLB (3) FTD (2)	PACS (12) CSS (5) Caregiver report (5) FMTCS (4) Revised CSS (2) GAIN (2) Meaning in caregiving scale (2) PAC questionnaire (1) Gains associated with caregiving (1) Personal gain (1) Caregiver esteem (1) Caregiving gratification scale (2) Strawbridge caregiving satisfaction scale (1) Appraisals of gain (1) CASI (1)	Cross-sectional (30) Measure validation (5) Longitudinal (5)	30 (11-38)
	No significant association	6					
	Related findings	8 Predicted and mediated variance. Direct relationship in Path model.					
Health (16)	Associated with	6	Spouse	AD (7)	Strawbridge caregiving	Cross-sectional	31.6

Caregiver well-being outcomes (N associations)	Findings	Number of studies/Details	Kin-relationship	Dementia diagnosis (N)	Positive aspects of caregiving measures (N)	Design employed (N)	Quality rating *M* (range)
	better health		Partner Child Child-in-law Sibling Niece Relative Friend Other	Mixed (2) Vascular (2) PD (1) HC (1)	satisfaction scale (4) PACS (3) FMTCS (2) CSS (2) BENEFIT (1) Meaning in caregiving scale (1) CASI (1) Appraisals of gain (1)	(14) Measure validation (2)	(24-38)
	No significant association	10					
Role strain (11)	Associated with lower role strain	4	Spouse Partner Child Child-in-law Sibling Grandchild Relative Other	AD (3) Vascular (3) DLB (1)	PACS (3) Satisfaction with caregiving role (2) Meaning in caregiving scale (2) FMTCS (1) Rewards of caregiving scale (1) Adapted Strawbridge caregiving satisfaction scale (1) Personal gain (1)	Cross-sectional (9) Measure validation (1) Longitudinal (1)	30.7 (24-35)
	No significant association	5					
	Related findings	2 Predicted variance and variance not predicted					
Satisfaction with life (4)	Associated with greater satisfaction with life	2	Spouse Partner Child Parent Sibling Relative Other	AD (3)	CSS (2) Gains associated with caregiving (1) PACS (1)	Cross-sectional (3) Measure validation (1)	32.8 (30-36)
	No significant association	1					
	Related findings	1 Predicted variance					
QoL/ HRQoL (9)	Associated with better QoL/HRQoL	6	Spouse Child Child-in-law Niece Relative	AD (5) Mixed (2)	PACS (4) Caregiver esteem (2) FMTCS (1) BENEFIT (1) Rewards of caregiving (1)	Cross-sectional (9)	32.2 (28-36)
	Associated with worse QoL/HRQoL	1					
	No significant	1					

Caregiver well-being outcomes (N associations)	Findings	Number of studies/Details	Kin-relationship	Dementia diagnosis (N)	Positive aspects of caregiving measures (N)	Design employed (N)	Quality rating *M* (range)
	association		Other				
	Related findings	1 Predicted variance					
Mental health (25)	Associated with better mental health	18	Spouse Partner Child Child-in-law Sibling Grandchild Cousin Niece Relative Friend Other	AD (18) Vascular (12) Mixed (4) DLB (2) FTD (1)	Caregiver report (6) Satisfaction with caregiving role (4) PACS (3) CSS (3) FMTCS (3) Appraisals of gain (2) Caregiver esteem (2) GAIN (1) Personal gain (1)	Cross sectional (15) Measure validation (1) Longitudinal (9)	29.8 (22-38)
	No significant association	5					
	Related findings	2 Predicted variance					
Personal strain/ stress (5)	Associated with less strain/stress	1	Spouse Partner Child Child-in-law Sibling Other	AD (2) Vascular (2) Mixed (1) DLB (1) PD (1) HC (1)	CSS (1) Personal gain (1) Adapted Strawbridge caregiving satisfaction scale (1) Strawbridge caregiving satisfaction scale (1) Caregiving rewards scale (1)	Cross sectional (4) Measure validation (1)	25.4 (11-37)
	Associated with more strain/stress	1					
	No significant association	3					
Competence/ self-efficacy (7)	Associated with greater competence/self-efficacy	4	Spouse Child Child-in-law Sibling Grandchild Relative Friend Other	AD (2) Vascular (1) DLB (1)	PACS (2) Meaning in caregiving scale (2) Revised CSS (1) GAIN (1) Personal gain (1)	Cross sectional (6) Measure validation (1)	29.1 (22-33)
	No significant association	2					
	Related findings	1 Predicted variance					

Abbreviations: AD- Alzheimer’s Disease; CASI- Carers’ Assessment of Satisfaction Index; CCS- Caregiving Satisfaction Scale; DLB- Dementia with Lewy Bodies; FMTCS- Finding Meaning Through Caregiving Scale; FTD- Fronto Temporal Dementia; GAIN- Gains in Alzheimer’s Care Instrument; HRQoL-Health-related Quality of Life; HC- Huntington’s Chorea; MICS- Meaning in Caregiving Scale; N- Number; PACQ- Positive Aspects of Caregiving Questionnaire; PACS- Positive Aspects of Caregiving Scale; PD- Parkinson’s Disease; QoL- Quality of Life

Supplementary Table 2. PRISMA checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	3-6
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	6
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	6
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	7
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	7
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	Supplementary document 1
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	6-8; Figure 1

Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	8
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	7; Supplementary document 2
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	8
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	8-9
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	9
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	19-20
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	N/A
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	Figure 1.
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	Table 2 and Supplementary table 1
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	Table 2
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	Supplementary table 1
Synthesis of results	21	Present the main results of the review. If meta-analyses are done, include for each, confidence intervals and measures of consistency	9-16
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	N/A

Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	N/A
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	16-17
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	19-20
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	16-22
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	N/A