Title:

Sense of Coherence, Burden and Mental Health in Caregiving: A Systematic Review and Meta-Analysis.

Running head:

Sense of coherence, burden and mental health in caregiving

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Dr Vasiliki Orgeta and Catalina López-Martínez were authors in two and one of the included studies.

Abstract

Background: Informal caregiving is associated with a number of negative effects on carers' physical and psychological well-being. The salutogenic theory argues that sense of coherence (SOC) is an important factor in psychological adjustment to stress. The main aim of this study was to systematically review current evidence on the association between SOC, burden and mental health outcomes in informal carers.

Method: A systematic search was carried out up to September 2017 in the following databases: PubMed, CINAHL (EBSCO), PsychInfo (OVID) and Scopus. Studies were included if they evaluated the relationship between sense of coherence and subjective caregiver burden and/or mental health outcomes, specifically symptoms of depression and anxiety. Meta-analyses were performed and subgroup analyses were carried out to explore if methodological factors influenced findings.

Results: Thirty-five studies were included in the meta-analysis, which provided 40 independent samples with 22 independent comparisons for subjective caregiver burden, 26 for symptoms of depression and 7 for symptoms of anxiety. Higher levels of SOC were associated with lower levels of subjective caregiver burden and better mental health outcomes. Publication bias did not change the estimate of the effect.

Limitations: Most of the studies included in this review were cross-sectional.

Conclusions: Findings suggest that SOC is an important determinant of carer well-being and may protect carers from high levels of psychological distress and caregiver burden.

Keywords: sense of coherence, caregiver burden, mental health, depression, anxiety

Introduction

Increasing life expectancy and current trends in population aging is leading to higher levels of dependency for older people through a rise of disabilities leading to increases in care and resources to meet care demands (OECD, 2013). Most of this care is delivered by unpaid carers generally women, referred to in the literature as informal carers (Del-Pino-Casado et al., 2011). Caring for a dependent person has negative effects on carers' physical and psychological health, and is associated with high levels of subjective burden, posing carers at increased risk of experiencing clinically significant symptoms of depression and anxiety (Cooper et al., 2007; van der Lee et al., 2014).

Subjective burden is a state characterised by fatigue, stress, and difficulties adjusting to the caregiving role. This state stems from a negative appraisal of the caregiving situation that can threaten the physical, psychological, and emotional health of carers (Kim et al., 2012; Zarit et al., 1980). Experiencing carer burden is often associated with outcomes such as anxiety and depression (Cooper et al., 2007). Anxiety includes feelings of worry and fear, and physical symptoms such as muscular tension and somatic symptoms whereas depressive symptoms are characterised by sad mood, loss of interest or joy in daily activities, fatigue, and excessive feelings of guilt and worthlessness (American Psychiatric Association, 2013; Goldberg et al., 1988).

Systematic reviews have shown prevalence rates of depression and anxiety of 40.2% and 21.4% (respectively) for stroke carers (Loh et al., 2017), and at 34.0% and 43.6 for carers of people living with dementia (Sallim et al., 2015). Studying the psychological determinants of caregiver burden and well-being is important for developing interventions to support carers and prevent psychological distress (Livingston et al., 2014).

High levels of sense of coherence (SOC) may protect individuals from stress and are associated with a reduced risk for various health problems (Jaracz et al., 2012). SOC comprises elements of comprehensibility (cognitive), manageability (instrumental) and meaningfulness (motivational) (Antonovsky, 1993), and refer to one's ability to understand a particular situation and use available resources effectively (material, psychological and social) allowing the individual to make use of adaptive coping strategies (Eriksson and

Lindstrom, 2007). As a result, individuals who endorse high levels of SOC view the world and their environment as more comprehensive, manageable, and meaningful (Lindstrom and Eriksson, 2005). SOC develops in young adulthood (Välimäki et al., 2009), as a result of learning rather than heredity (Kuroda et al., 2007), and although is considered to remain relatively stable across the life span, studies show that it increases with age (Lindstrom and Eriksson, 2005).

There are currently no systematic reviews of the literature quantifying the association of SOC with caregiver burden and mental health outcomes. The review by Zauszniewski et al. (2015) was narrative; focusing on carers of adults with serious mental illness, and therefore did not include all caregiving groups. A further review by (Rigby et al., 2009) included studies on carers of stroke survivors and did not include a meta-analysis. In this review we aimed to extend the available evidence base on SOC, caregiver burden and mental health outcomes across all caregiving groups; a secondary aim was to estimate the size of the association and report on the quality of the evidence.

Method

We searched major healthcare databases: PubMed, CINAHL (EBSCO), PsychInfo (OVID) and Scopus, up until September 2017. We followed the MOOSE (Stroup et al., 2000) and PRISMA statements (Moher et al., 2009), when reporting findings of this review. Our search included the following terms: caregiver (or carer), sense of coherence, salutogenesis, and Antonovsky, with no additional filters used. We consulted relevant authors for obtaining grey literature and unpublished studies, and searched the reference lists of all relevant publications and reviews.

Inclusion criteria

For individual studies, the following inclusion criteria were used: (1) original articles (2) reporting on the statistical association of SOC and subjective burden, depressive and/or anxiety symptoms (3) those that reported a correlation coefficient, and (4) included samples of informal carers (family, friends, community members or volunteers). Two independent reviewers (RdPC and AEM) selected studies with disagreements resolved by consensus.

Data extraction and synthesis

Two independent reviewers (RdPC and AEM) extracted the population characteristics and effect estimates of each study using a standardised data extraction form. Disagreements were resolved by consensus. The effect measure used to compute pooled estimates was the correlation coefficient. In repeated measures studies were correlations referred to the same time point, only the first correlation was selected in order to guarantee independence of comparisons (Higgins and Green, 2008). For computing correlation coefficients, we used the correction for reliability (internal consistency) following the formula proposed by Hunter and Schmidt: $\frac{r}{\sqrt{\alpha_x}\times\sqrt{\alpha_y}}$, r being the correlation coefficient and α_x and α_y the internal consistency coefficients of the independent and dependent variable, respectively.

Quality assessment

Basing on the recommendations of Boyle (1998) and Viswanathan, Berkman, Dryden, & Hartling (2013), we used the following criteria for assessing methodological quality of the individual studies: (1) representative sampling (probabilistic sampling), (2) reliability and validity of measures: content validity and internal consistency in the target or similar population, (3) control of confounding factors: at least one measure of objective burden must be controlled for and the variation of the point estimate must be less than 15%, (4) for longitudinal studies: (4.1) follow-up of at least six months and (4.2) rate of follow-up of at least 80% of the original population taking part. Two reviewers (RdPC and CLM) assessed quality of included studies independently.

In regards to confounders, we decided to control for objective burden because this construct is the main determinant of subjective burden, and of symptoms of depression and anxiety (Cooper et al., 2007; van der Lee et al., 2014). Objective burden encompasses patients’ needs (i.e. functional capacity, cognitive impairment and behavioural problems) and intensity of care (Aneshensel et al., 1995). Because measures of previous dimensions of objective burden are highly correlated (Pinquart and Sorensen, 2003), we controlled for at least one of these. We considered confounders being controlled in studies by taking into account those related to the design and/or analysis (i.e. matching, stratification, interaction terms, multivariate analysis, or other statistical adjustment such as instrumental variables) (Viswanathan et al., 2013). In cases of statistical adjustment, we considered no confounding bias to be present when the variation of the point estimate was less than 10% (Rothman et al., 2008).

Following the recommendations of (Meader et al., 2014), and the Grading of Recommendations Assessment, Development and Evaluation (GRADE) system (Atkins et al., 2004), inconsistency, imprecision and risk of publication bias were also evaluated. Inconsistency was evaluated through heterogeneity of findings in individual studies and imprecision through the number of included studies (large: >10 studies, moderate: 5-10 studies and small: < 5 studies) and the median sample size (high: > 300 participants, intermediate: 100-300 participants and low: < 100 participants). Publication bias was assessed by analysing the funnel plot and statistical tests (see Analyses section).

Analyses

We used a random-effects model for estimating pooled effects and their 95% confidence intervals (CI), in order to improve generalisation of study findings (Cooper et al., 2009). The Q test was used for heterogeneity analysis, calculating the degree of inconsistency (I^2) (Higgins et al., 2002). For publication bias, we used the Begg test (Begg and Mazumdar, 1994), the Egger test (Egger et al., 1997) and the pooled effect; estimated by taking into account possible publication bias by the Trim and Fill method (Duval and Tweedie, 2000). We used several approaches to analyse publication bias in order to look for concordant results that may strengthen conclusions (Guyatt et al., 2011). Sensitivity analyses were carried out to assess the contribution of studies to the pooled estimate in each meta-analysis (leave-one-out method; (Cooper et al., 2009), and to assess whether study design or other quality parameter influenced results (sub-group analyses; (Cooper et al., 2009). Comprehensive Meta-analysis 3.3 was used for the calculations.

Results

A total of 447 studies were retrieved from databases and 8 studies were identified from searching references of included articles (7 studies) or contacting authors (1 unpublished study) (see Figure 1). After removing duplicates, 231 records remained to be screened. A total of 143 records were excluded as not being relevant and 50 studies did not meet the inclusion criteria. After removing 3 duplicate articles (Gustavsson-Lilius et al., 2007; Jaracz et al., 2012; Zauszniewski et al., 2009), 35 studies remained to be screened for inclusion.

Of these 34 were published studies (Andrén and Elmståhl, 2008; Bias, 1998; Bowe, 2003; Caap-Ahlgren and Dehlin, 2002; Coe et al., 1991; Chumbler et al., 2004; Dejo Vásquez, 2007; Fan et al., 2014; Forsberg-Warleby et al., 2002; Gallagher et al., 1994; Götze et al., 2015; Gustavsson-Lilius et al., 2012; Hiyoshi-Taniguchi et al., 2014; Hsiao and Tsai, 2015; Jaracz et al., 2015; Jaracz et al., 2014; Lo Sterzo and Orgeta, 2017; Matsushita et al., 2014; Michailidou et al., 2013; Olsson and Hwang, 2002; Orgeta and Lo Sterzo, 2013; Perrin et al., 2008; Shroff, 2014; Suresky et al., 2014; Sutter et al., 2016; Tang et al., 2013; Thompson et al., 2004; Trujillo et al., 2016; Truong, 2015; Välimäki et al., 2009; Van Puymbroeck et al., 2008; Weimand et al., 2010; Wu et al., 2015; Zauszniewski and Suresky, 2010), and one was unpublished (López-Martínez, 2016). Across the 35 included studies there were 40 independent samples with 22 independent comparisons of SOC with subjective burden, 26 with depressive symptoms and 7 with symptoms of anxiety. The main characteristics of included studies can be seen in Table 1. A total of 29 studies were cross-sectional, 5 were repeated measures studies and one was a case-control study.

Most studies used Antonovsky's SOC scale to measure SOC, with 20 studies incorporating the 13-item version, 12 the 29-item version and 2 studies a modified version developed by the authors. The study by Shroff (2014) used the "Family Coping Coherence Index". Most were written in English, two in Spanish, one in Chinese, one in Greek and one in German. As can be seen in Table 2, all studies but 3 had non-probabilistic samples, and all reported estimates of validity and reliability of measures used. A total of 9 studies reported controlling for confounders.

SOC and subjective burden

Twenty studies with 22 samples and independent comparisons examined the relationship between SOC and subjective burden (see Table 1). These studies were published between 1994 and 2016. Most were cross-sectional with the exception of three which were repeated measures studies, even though the correlations reported referred to the same time point. All but three had non-probabilistic sampling. The main patient groups were people with dementia (seven studies), stroke survivors (five studies) and frail older people (four studies).

The forest plot of the meta-analysis is presented in Figure 2. The pooled effect ($\bar{r}$ = -0.53; 95% CI = -0.60, -0.46; N= 3,120) showed a moderate negative association between SOC and subjective burden. All the effects of the individual studies had the same direction and were statistically significant. Most of the studies had confidence intervals that overlapped with each other. The pooled effect size estimate is considered precise because of the number of included samples (22) and median sample size (135.7). Heterogeneity of individual studies was generally low (Q = 21.28, degrees of freedom [df] = 21, p = 0.41, I^2 = 1.3%). Sensitivity analysis (one study removed) yield variations in the pooled estimate under 3%. The funnel plot (see Supplementary Figure 1) seemed somewhat symmetric and the results of the Begg (p = 0.43) and Egger Test (p = 0.51) indicated that publication bias was not present. This was in line with the pooled estimate corrected by the Trim and Fill method varying only by 4% ($\bar{r}$ = -0.51).

No differences were found between studies that controlled ($\bar{r}$ = -0.51; 95% CI = -0.58, -0.43; 9 samples) or did not control for objective burden ($\bar{r}$ = -0.55; 95% CI = -0.65, -0.43; 13 samples). The pooled estimate was greater for studies with non-probabilistic samples ($\bar{r}$ = -0.55; 95% CI = -0.61, -0.47; 19 samples) compared to those that did use probabilistic sampling ($\bar{r}$ = -0.44; 95% CI = -0.61, -0.23; three samples) but the difference was not significant.

SOC and depressive symptoms

A total of 22 studies with 26 samples and independent comparisons were included to quantify the association between SOC and depressive symptoms (see Table 1). Year of publication ranged from 1998 to 2017. All studies were cross-sectional except from four (three were repeated measures studies and one was a case-control study). Among the repeated measures studies, two had correlations referred to at the same time point and one had longitudinal measures with correlations referred to different time points. All studies except one had non-probabilistic sampling. Seven studies included carers of people with dementia, seven studies recruited carers of persons with stroke, and three studies carers of people with cancer.

The forest plot of the meta-analysis is presented in Figure 2. The pooled effect ($\bar{r}$ = -0.69; 95% CI= -0.75, -0.63; N= 3,216) showed a moderate to large negative association between SOC and depressive symptoms. All individual studies reported effects on the same direction and were statistically significant, with substantial overlap overall on confidence intervals. The results of the meta-analysis were considered precise (included samples = 26; median sample size = 110.9). There was low heterogeneity overall (Q = 25.31, df = 25, p = 0.40), I^2 = 1.2%). Sensitivity analysis (one study removed) showed variations in the pooled estimate under 1.5%. Inspection of the funnel plot (see Supplementary Figure 2) showed no presence of bias, which was confirmed by the results of the Begg (p = 0.68), and Egger Test (p = 0.39), and the pooled estimate corrected by the Trim and Fill method ($\bar{r}$ = -0.69).

The pooled estimate for studies controlling for confounders ($\bar{r}$ = -0.77; 95% CI = -0.81, -0.72; 5 samples) was higher compared to those not controlling for confounders ($\bar{r}$ = -0.67; 95% CI = -0.74, -0.59; 3 samples), but the difference was not significant. We were not able to perform sensitivity analysis by type of sampling because there was only one study with probabilistic sampling.

In regards to effect of study design, studies with a longitudinal design (case control study and repeated measures study with correlations referred to different time points) showed a greater pooled effect ($\bar{r}$ = -0.78; 95% CI = -0.85, -0.69; five samples) than cross-sectional and repeated measures studies with cross-sectional correlations ($\bar{r}$ = -0.67; 95% CI = -0.73, -0.59; 21 samples); but this difference was not significant.

SOC and anxiety symptoms

Seven studies with seven samples and independent comparisons were pooled to quantify the association between SOC and anxiety (see Table 1). Publication year for studies ranged from 2002 to 2017. All studies except two (repeated measures studies) were cross-sectional. One of these had correlations that referred to the same time point and the other had correlations referring to different time points. All studies but one had non-probabilistic sampling. Three of the studies included carers of people with dementia, two studies carers of stroke survivors, one study carers of relatives with cancer, and the final study carers of frail older people.

The forest plot of the meta-analysis is presented in Figure 2. The pooled effect ($\bar{r}$ = -0.60; 95% CI= -0.70, -0.47; N= 940) showed a moderate to large negative association between SOC and anxiety. All individual studies reported effects on the same direction and were statistically significant. Confidence intervals across included studies generally overlapped. The results of the meta-analyses were considered precise due to the number of included samples (seven) and their median sample size (117.5). Low heterogeneity was found across individual studies (Q = 6.73, df = 6, p = 0.35, I^2 = 10.9%). Sensitivity analysis (one study removed) showed variations in the pooled estimate under 6.4%. The funnel plot (see Supplementary Figure 3) appeared somewhat asymmetric; there were no outliers, and small studies had no bigger effect sizes than larger studies. Similarly the results of the Begg (p = 0.76) and Egger Test (p = 0.66) confirmed that there was no publication bias present, and the pooled estimate corrected by the Trim and Fill method did not vary ($\bar{r}$ = -0.60). We were unable to perform sensitivity analysis on effects of sampling, controlling for confounders, due to the small number of studies overall (there was only one study using probabilistic sampling; one study controlling for confounders; one longitudinal study).

Discussion

This is the first systematic review and meta-analysis to examine the relationship between SOC, subjective burden and mental health outcomes in caregiving populations. Our review is the first comprehensive systematic review of the worldwide literature on the relationship between SOC, subjective burden and mental health outcomes in informal carers. Our quantitative syntheses provide evidence that sense of coherence has a moderate negative impact on subjective caregiver burden, and a moderate to large effect on symptoms of depression and anxiety. To our knowledge this is the first systematic review providing a quantitative synthesis of the evidence, which forms an important foundation for future research and the development of interventions that aim to increase sense of coherence and coping in caregiving populations.

Our findings are consistent with the work of Erikson and Lindstrom (2006) where SOC is related to better health and quality of life outcomes. Several researchers have argued that SOC allows better management of stressful events through appraising these as more comprehensible, meaningful and manageable (Lindstrom and Eriksson, 2005). Our meta-analysis findings show that there is a consistent negative association between subjective caregiver burden and SOC, with the effect size interpreted as moderate.

High levels of subjective burden involves the perception and evaluation of the caregiving situation and role as stressful (Roche et al., 2015). Therefore high SOC in carers or interventions that assist carers in viewing caring duties as more manageable are likely to mediate subjective burden. For symptoms of anxiety and depression we found that the effect size of the association was moderate to large, indicating that SOC mediates psychological distress in family carers and should therefore be an important target for future interventions.

The findings of our meta-analyses performed were reasonably accurate, valid, and robust for subjective burden and depressive symptoms. Overall there was a high number of included studies, with low levels of heterogeneity observed, and low risk associated with publication bias. We were also able to perform sensitivity analysis for these outcomes indicating that generally the effect estimates are relatively stable. However, our analyses for the association of anxiety symptoms and SOC are limited, primarily due to the small number of studies included. Overall there was a low risk of classification bias across all studies due to the use of validated and reliable measures. However most of the studies used non-probabilistic samples and some did not control for confounders.

Given that the majority of studies were cross-sectional there are potential threats to the validity of our meta-analyses. This means that the association of SOC and subjective burden, and symptoms of anxiety and depression could be due to ‘reverse causation’. Given that all studies included in the meta-analysis of subjective burden were cross-sectional or repeated measures studies no causal relationship can be inferred. However, the low levels of heterogeneity in the effects of individual studies, and the overlap of the CIs indicate that the effects are to some extent stable to potential confounders. For example, there were no differences between studies with probabilistic and non-probabilistic sampling, limiting the potential threat to external validity.

Studies reporting on the association between SOC, depressive and anxiety symptoms were mostly cross-sectional, with only two high-quality longitudinal studies for depressive symptoms and one for symptoms of anxiety. We found no statistical differences in the pool estimates of these type of studies for depressive symptoms. For symptoms of anxiety, the effect of the longitudinal design was similar to the pooled effect indicating that type of design at least on the available evidence to date does not influence the size of the association with SOC.

One of the main difficulties in systematic reviews of observational studies is the control for confounders (Viswanathan et al., 2013). In the present review, we applied specific strategies for evaluating this. For example, we demonstrated that controlling for objective burden in individual studies did not affect the results of our meta-analyses. Thus, taking into account available evidence our review concludes that overall experiencing low levels of subjective burden, depression and anxiety symptoms are associated with higher levels of SOC across several caregiving populations. Our findings allow us therefore to conclude that SOC is an important determinant of carer well-being, protecting carers from high levels of psychological distress associated with burden, low mood and anxiety.

More research is needed however in order to understand the association between SOC and subjective burden, depression and anxiety symptoms, and identifying mediators of these relationships. We were unable to examine for which subgroups SOC may have a disproportionate effect on mental health and for which caregiving outcomes. We were also not able to test which demographic characteristics may interact with SOC to reduce or increase subjective burden or psychological distress. The majority of studies were conducted in high-income countries limiting our conclusions. Although SOC is conceptualised as a cross-cultural concept it will be important to study effects of culture and how availability of resources may differ for carers across different cultures and settings. For example, levels of SOC may differ across groups depending on cultural values which may exert different effects on stress reactions for carers. Our use of subgroup analysis was informative, but it was limited by the small number of studies included. Longitudinal studies are required to demonstrate causal-effect relationships.

Knowledge of the impact of sense of coherence on carer psychological well-being is important clinically. Clinicians should be aware that low sense of coherence in carers may signal psychological distress such as experiencing clinically significant symptoms of anxiety and depression. Screening questions by clinicians can help identify carers with low SOC and those that may need access to specialist psychological support or enhanced support to cope with their caregiving role.

In recent years there has been increasing interest on how to strengthen SOC (Yamazaki et al., 2011) for patient and informal carers with several studies showing that SOC increases through interventions that focus on coping (Lim and Han, 2013), problem solving (Odajima et al., 2017), cognitive therapy (Malm et al., 2018) or lifestyle interventions (Forsberg et al., 2010); although informative large scale randomised controlled trial studies of clinical effectiveness are needed.

To conclude our review shows that SOC is associated with important outcomes for carers across different populations. SOC has a consistent association with caregiver burden and psychological distress specifically symptoms of anxiety and depression. Our meta-analysis shows that SOC-strengthening interventions may reduce the risk of emergence of depression and anxiety symptoms in carers and protect their well-being and urges for further research to investigate the effects of these interventions.

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Table 1. Main characteristics of included studies with their independent samples.

	Sample size	Design	Care recipients	Outcomes
Andrén & Elmståhl, 2008	130	CS	Dementia	Subjective burden
Bias, 1998	112	CS	Dementia	Depressive symptoms
Bowe, 2003	68	CS	Cancer	Subjective burden, depressive symptoms
Caap-Ahlgren & Dehlin, 2002	65	RM1	Parkinson	Subjective burden
Chumbler, Rittman, Van Puymbroeck, Vogel, & Qin, 2004	102	CS	Stroke	Subjective burden, depressive symptoms
Coe, Miller, & Flaherty, 1991	148	CS	Chronic	Depressive symptoms
Dejo Vasquez, 2007	74	CS	Frail older people and people with dementia	Subjective burden
Fan, Mei-BIH, Lin, Bai, & Wei, 2014	70	CS	Mental illness	Depressive symptoms
Forsberg-Warleby, Moller, & Blomstrand, 2002	76	CS	Stroke	Depressive symptoms, anxiety symptoms
Gallagher, Wagenfeld, Baro, & Haepers, 1994	71	CS	Frail older people	Subjective burden
	55		Dementia	Subjective burden
Götze, Brähler, Gansera, Schnabel, & Köhler, 2015	105	CS	Cancer	Subjective burden
Gustavsson-Lilius, 2012	147	RM2	Cancer	Depressive symptoms, anxiety symptoms
Hiyoshi-Taniguchi, Becker, & Kinoshita, 2014	177	CS	Frail older people	Subjective burden
Hsiao & Tsai, 2015	137	CS	Mental Illness	Subjective burden
Jaracz, Grabowska-Fudala, Górna, & Kozubski, 2014	150	RM1	Stroke	Subjective burden, depressive symptoms, anxiety symptoms
Jaracz et al., 2015	88	RM1	Stroke	Subjective burden
Lo Sterzo & Orgeta, 2017	155	CS	Dementia	Depressive symptoms, anxiety symptoms
López Martínez 2016[a]	132	CS	Frail older people	Subjective burden, depressive symptoms, anxiety symptoms
Matsushita et al., 2014	274	CS	Dementia	Subjective burden
Michailidou, Anagnostopoulos, Vemmos, & Michailidis, 2013	106	CS	Stroke	Depressive symptoms

Notes: [a] grey literature.
Abbreviations: CS: cross-sectional studies; RM: repeated measures studies.

Table 1. Main characteristics of included studies with their independent samples (continued).

	Sample size	Design	Care recipients	Outcomes
Olsson & Hwang, 2002	62	C&C	Autism (children)	Depressive symptoms
	47		Autism (children)	Depressive symptoms
	144		Other disabilities (children)	Depressive symptoms
	115		Other disabilities (children)	Depressive symptoms
Orgeta & Lo Sterzo, 2013	170	CS	Dementia	Depressive symptoms, anxiety symptoms
Perrin, Heesacker, Stidham, Rittman, & Gonzalez-Rothi, 2008	135	CS	Stroke	Subjective burden, depressive symptoms
Shroff, 2014	83	CS	Dementia	Subjective burden
Suresky et al., 2014	60	CS	Mental illness	Subjective burden
Sutter et al., 2016	127	CS	Dementia	Subjective burden, depressive symptoms
Tang et al., 2013	621	CS	Cancer	Subjective burden, depressive symptoms
Thompson et al., 2004	61	CS	Dementia	Depressive symptoms
Trujillo et al., 2016	110	CS	Dementia	Depressive symptoms, anxiety symptoms
Truong, 2015	153	CS	Dementia	Subjective burden
Välimäki et al., 2009	170	CS	Dementia	Depressive symptoms
Van Puymbroeck, Hinojosa, & Rittman, 2008	87	CS	Stroke	Subjective burden, depressive symptoms
Weimand, Hedelin, Sällström, & Hall-Lord, 2010	226	CS	Mental illness	Subjective burden
Wu, Lee, Su, & Pai, 2015	41	MR1	Stroke	Depressive symptoms
Zauszniewski & Suresky, 2010	60	CS	Mental illness	Depressive symptoms

Abbreviations: CS: cross-sectional studies; RM: repeated measures studies; C&C: case-control studies.

Table 2: Methodological quality assessment of individual studies.

	C1	C2	C3	C4.1	C4.2
Andrén & Elmståhl, 2008	–	+	–	N/A	N/A
Bias, 1998	–	+	–	N/A	N/A
Bowe, 2003	–	+	–	N/A	N/A
Caap-Ahlgren & Dehlin, 2002	–	+	–	N/A	N/A
Chumbler, Rittman, Van Puymbroeck, Vogel, & Qin, 2004	–	+	+	N/A	N/A
Coe, Miller, & Flaherty, 1991	–	+	–	N/A	N/A
Dejo Vasquez, 2007	–	+	+	N/A	N/A
Fan, Mei-BIH, Lin, Bai, & Wei, 2014	–	+	–	N/A	N/A
Forsberg-Warleby, Moller, & Blomstrand, 2002	–	+	–	N/A	N/A
Gallagher, Wagenfeld, Baro, & Haepers, 1994	–	+	+	N/A	N/A
Götze, Brähler, Gansera, Schnabel, & Köhler, 2015	–	+	–	N/A	N/A
Gustavsson-Lilius, 2012	–	+	–	+	+
Hiyoshi-Taniguchi, Becker, & Kinoshita, 2014	–	+	+	N/A	N/A
Hsiao & Tsai, 2015	–	+	–	N/A	N/A
Jaracz, Grabowska-Fudala, Górna, & Kozubski, 2014	–	+	–	N/A	N/A
Jaracz et al., 2015	–	+	–	N/A	N/A
Lo Sterzo & Orgeta, 2017	–	+	–	N/A	N/A
López Martínez 2016[a]	+	+	+	N/A	N/A
Matsushita et al., 2014	+	+	+	N/A	N/A
Michailidou, Anagnostopoulos, Vemmos, & Michailidis, 2013	–	+	–	N/A	N/A
Olsson & Hwang, 2002	–	+	–	?	+
Orgeta & Lo Sterzo, 2013	–	+	–	N/A	N/A
Perrin, Heesacker, Stidham, Rittman, & Gonzalez-Rothi, 2008	–	+	–	N/A	N/A
Shroff, 2014	–	+	–	N/A	N/A
Suresky et al., 2014	–	+	–	N/A	N/A
Sutter et al., 2016	–	+	–	N/A	N/A
Tang et al., 2013	–	+	+	N/A	N/A
Thompson et al., 2004	–	+	–	N/A	N/A
Trujillo et al., 2016	–	+	–	N/A	N/A
Truong, 2015	+	+	–	N/A	N/A
Välimäki et al., 2009	–	+	+	N/A	N/A
Van Puymbroeck, Hinojosa, & Rittman, 2008	–	+	+	N/A	N/A
Weimand, Hedelin, Sällström, & Hall-Lord, 2010	–	+	–	N/A	N/A
Wu, Lee, Su, & Pai, 2015	–	+	–	N/A	N/A
Zauszniewski & Suresky, 2010	–	+	–	N/A	N/A

Notes: (+) characteristic is present; (-) characteristic is absent; (?) there is not enough information to assess; [a] grey literature.

Abbreviations: C1: representative sampling (probabilistic sampling); C2: reliability and validity of the measures; C3: control of confounding factors; for longitudinal studies: C4.1: follow-up; C4.2: attrition.

Figure 1. PRISMA flow diagram of the review process.

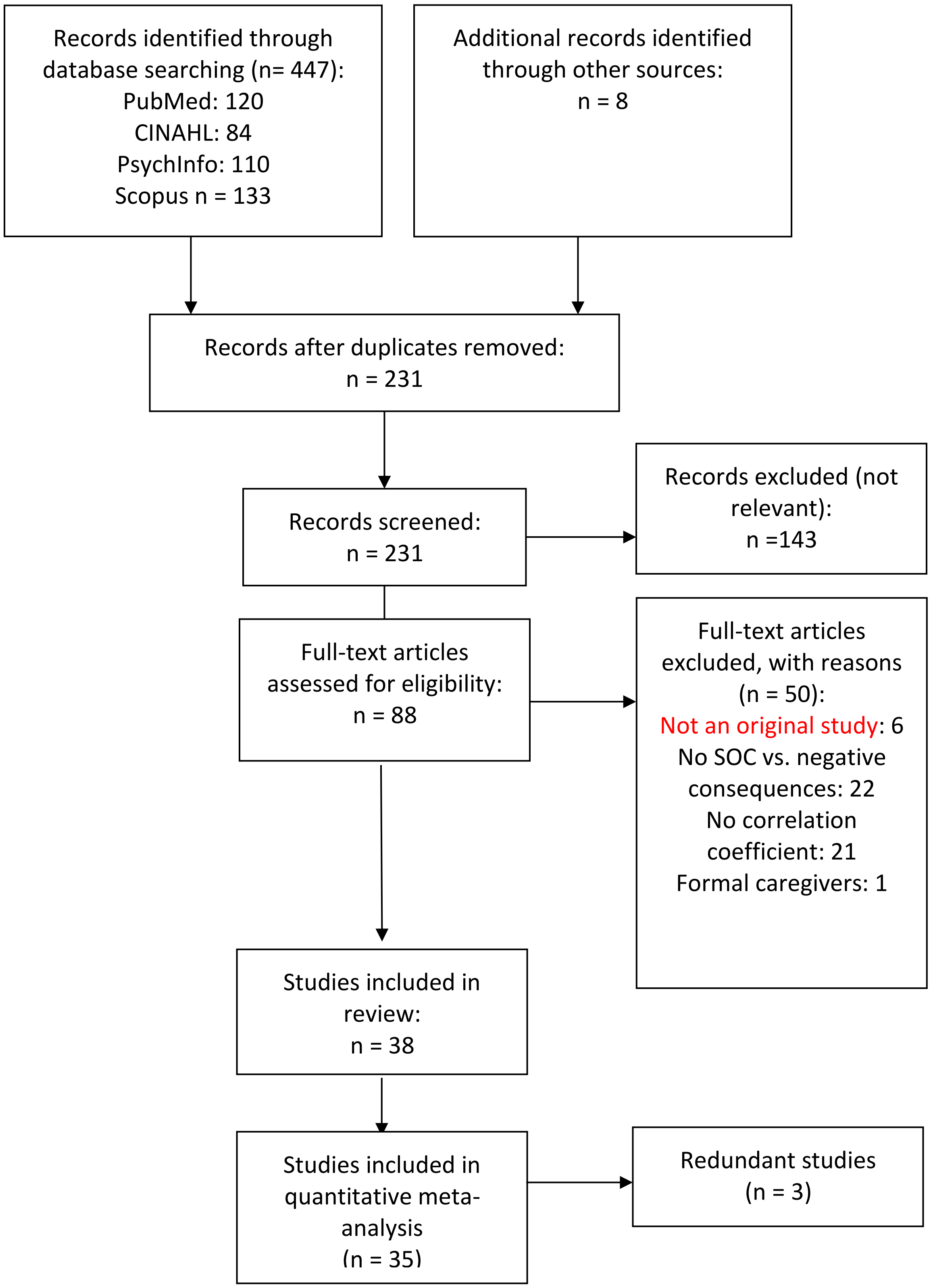
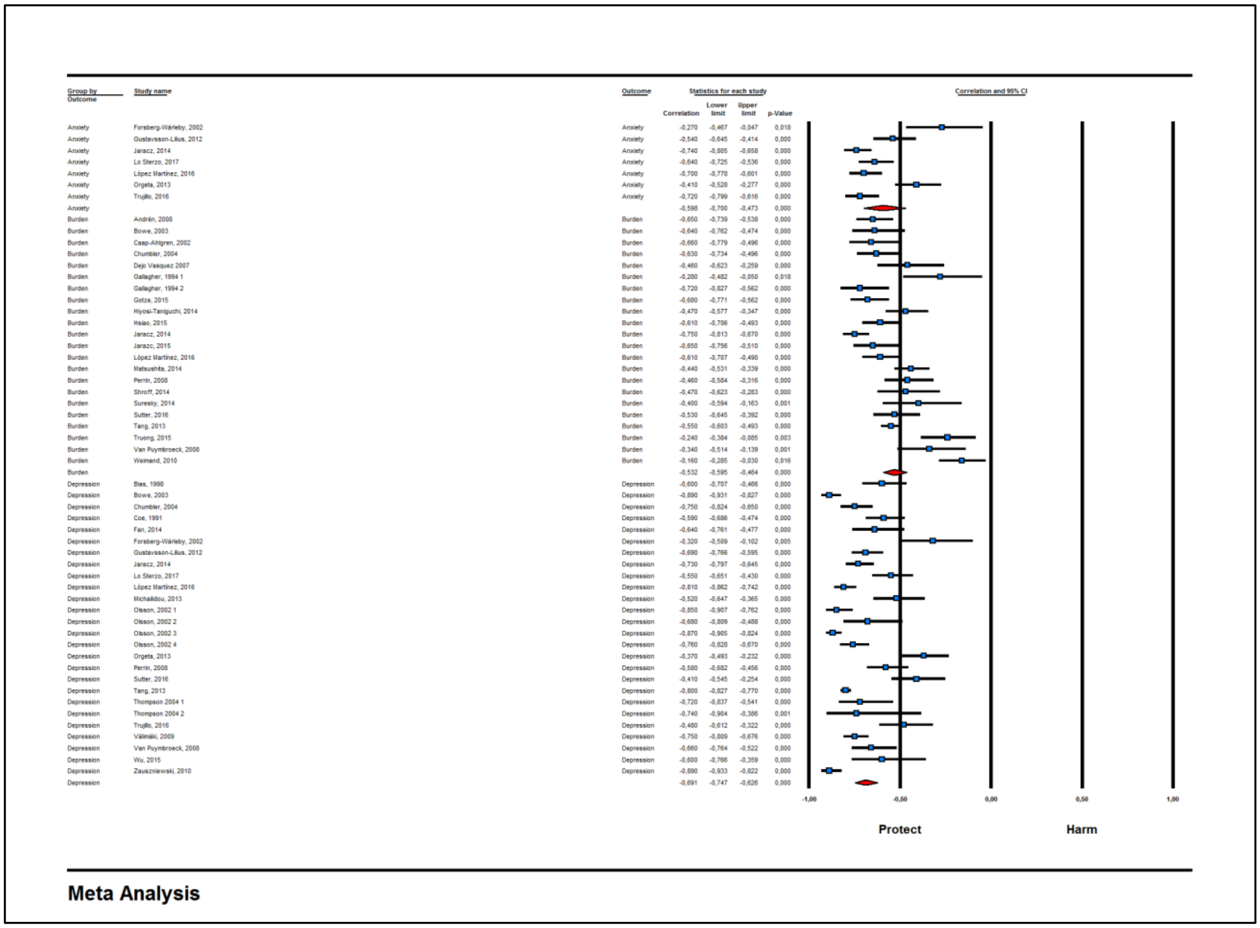
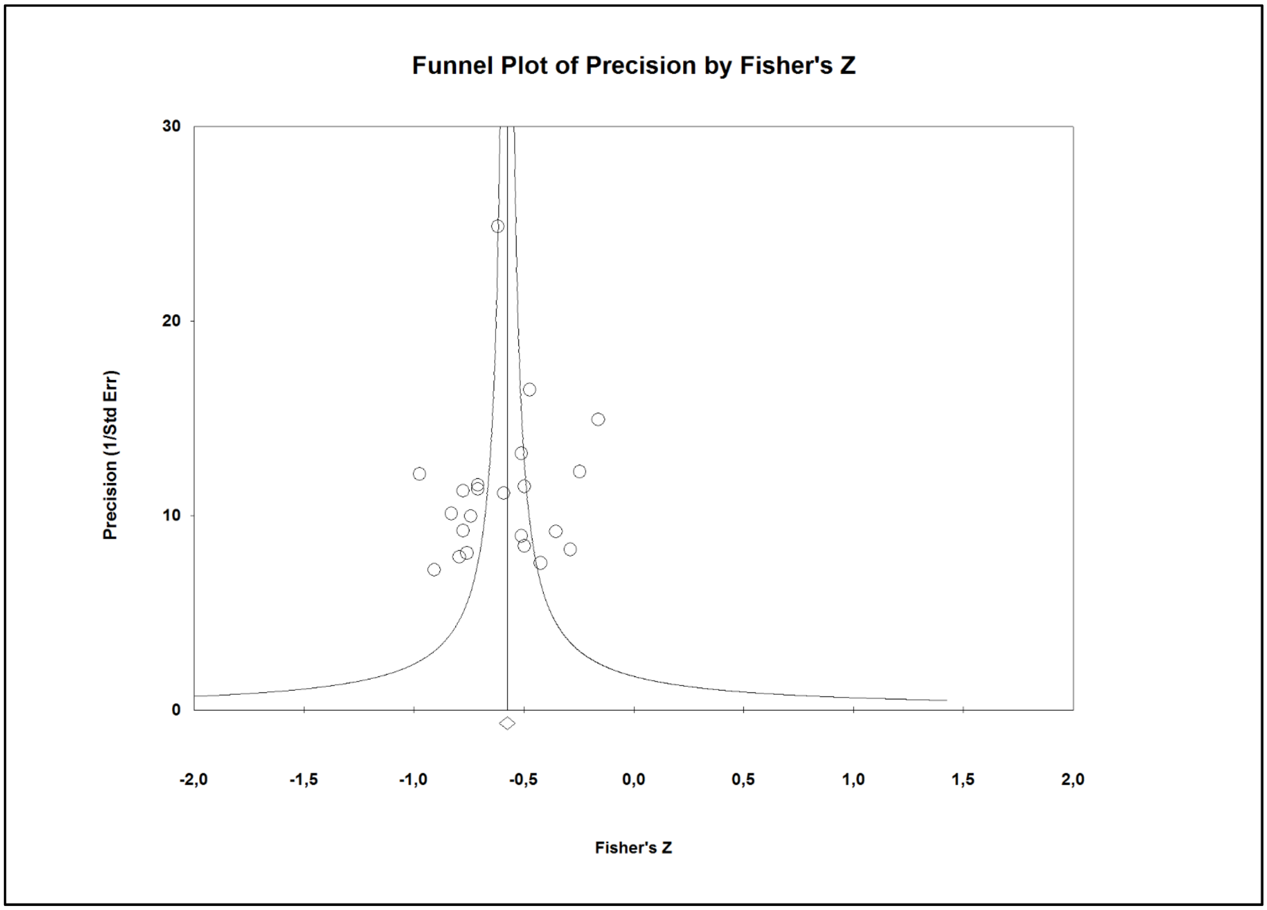


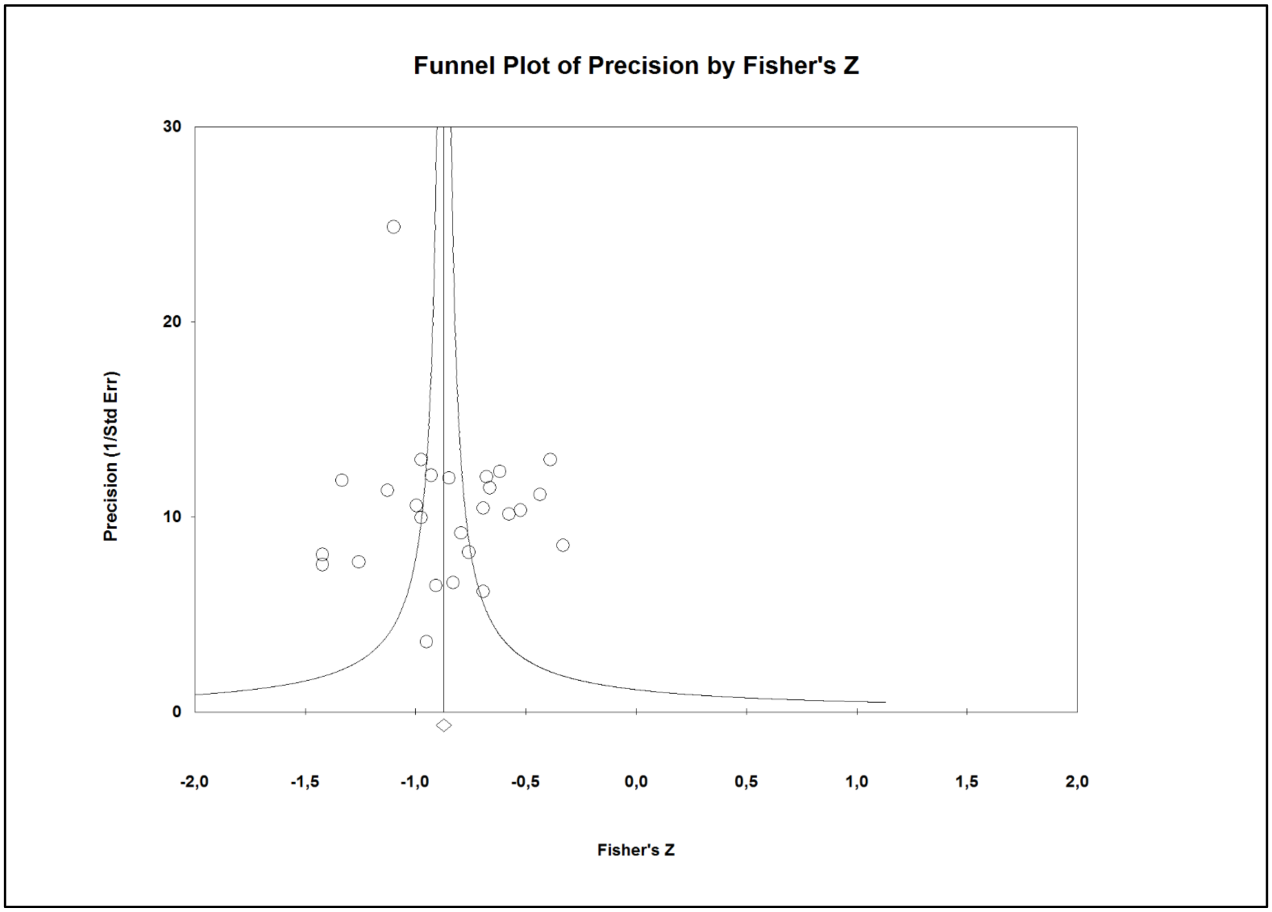
Figure 2. Forest plot for SOC and subjective burden, depressive symptoms and anxiety.



Supplementary Figure 1. Funnel plot for SOC and subjective burden.



Supplementary Figure 2. Funnel plot for SOC and depressive symptoms.



Supplementary Figure 3. Funnel plot for SOC and anxiety symptoms.