Doctorate in Clinical Psychology

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A systematic review of the predictors of carer burden in caregivers of children with chronic conditions

and

The relationship between carer burden, self-compassion, psychological flexibility and wellbeing in caregivers of children with chronic conditions

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Research Portfolio Abstract

Background: Caregivers of children with chronic conditions have been shown to experience higher levels of carer burden, which has been linked to increased mental health difficulties (such as anxiety and depression) and lower wellbeing. Evidence suggests that there are a number of factors which may act as predictors of carer burden in this population, however further research is needed to update the evidence base. A number of studies suggest a role for self-compassion and psychological flexibility as potential predictors of the relationship between carer burden and caregiver wellbeing. Research Article 1 synthesised findings regarding predictors of carer burden in caregivers of children with chronic conditions as an update from a review completed in 2012. Research Article 2 examined whether carer burden, self-compassion, psychological flexibility predict wellbeing in caregivers of children with chronic conditions.

Design: In Research Article 1 the authors conducted a systematic search using the electronic databases PsychINFO, MEDLINE, EMBASE, Cumulative Index to Nursing and Allied Health Literature as well as examining grey literature and reference lists of included studies. Findings were summarised using a narrative synthesis approach. In Research Article 2 a cross sectional design was used. Participants (*N* = 205) were recruited via social media, and completed an online survey encompassing demographic information and measures of burden (ZBI), self-compassion (SCS), psychological inflexibility (AAQ-II), anxiety (GAD-7), depression (PHQ-9) and quality of life (QoLS).

Results Preliminary results suggest that caregiver mental health difficulties, marital conflict and other stressors, high hours of caregiving, condition severity and family income/loss of work predict increased carer burden. Probable protective factors included good family functioning, social support, caregiver coping, family centred care and quality of life. Results from Research Article 2 showed that in combination carer burden, self-compassion and psychological inflexibility all significantly predicted anxiety, depression and quality of life with large effect sizes. Carer burden was found to be a predictor of higher anxiety and lower quality of life but did not predict depression. Psychological inflexibility predicted higher anxiety and depression scores and lower quality of life. Self-compassion predicted lower anxiety and depression scores and higher quality of life.

Conclusion: Overall the two studies highlight the important of supporting caregivers of children with chronic conditions, and the need for further high quality research in this population. Preliminary findings regarding potential predictor variables need to be further examined in a larger sample using standardised measures of carer burden. Carer burden, psychological inflexibility and self-compassion appear to be important targets for intervention to improve wellbeing in caregivers of children with chronic conditions. Future research is needed to examine how these predictors could be targeted by interventions to support caregiver wellbeing.

Lay Summary

In the summary below, the term carer or caregiver refers to anyone who takes care of a child with a chronic condition unpaid (including parents, foster families and family members). Previous studies show a link between experiencing stress as part of the caring role and lower caregiver wellbeing. This study explored whether any specific characteristics of the caregiver, child or condition might be related to increased stress in caregivers of children with chronic conditions by looking at research studies in the last 8 years. Preliminary results suggest that caregiver mental health difficulties, marital conflict and other stressors, high hours of caregiving, condition severity and family income/loss of work predict increased carer burden. Probable protective factors included good family functioning, social support, caregiver coping, family centred care and quality of life. Support should be made available to all caregivers, and clinicians should examine who might be more at risk of experiencing increased stress as part of their caring role in order to intervene at early on and prevent caregivers from experiencing increased stress and lower wellbeing. However there are a number of factors which are likely to affect this, and so further research is needed. This study also looked at whether carer burden (stress experienced through caring role), self-compassion (being kind to yourself) and psychological flexibility (being adaptable to change) predicted anxiety, depression and quality of life in caregivers. Results showed that having higher levels of caregiver stress was correlated with worse wellbeing. Having more self-compassion and psychological flexibility were related to less stress and better wellbeing in caregivers. Psychological flexibility and self-compassion were linked to less carer stress. Carer stress was found to be a predictor of higher anxiety and lower quality of life but did not predict depression. Psychological inflexibility (not being adaptable to change) predicted higher anxiety and depression scores and lower quality of life. Self-compassion predicted lower anxiety and depression scores and higher quality of life. More research is needed to help build interventions to support caregivers of children with chronic conditions.

Chapter 1. Systematic Review

A systematic review of the predictors of carer burden in caregivers of children with chronic conditions

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Abstract

Aim: This review synthesised findings regarding predictors of carer burden in caregivers of children with chronic conditions. High levels of carer burden have been found in this population, and a previous review examined studies prior to 2012. This review updates the evidence base regarding potential predictors in order to begin informing the development potential early identification and support tools for caregivers. **Design:** A systematic search was conducted using the electronic databases PsychINFO, MEDLINE, EMBASE, Cumulative Index to Nursing and Allied Health Literature, grey literature and reference lists of included studies. Findings were summarised using a narrative synthesis approach. **Results:** Preliminary results suggest that caregiver mental health difficulties, marital conflict and other stressors, high hours of caregiving, condition severity and family income/loss of work predict increased carer burden. Probable protective factors included good family functioning, social support, caregiver coping, family centred care and quality of life. **Conclusion**: The review highlights preliminary findings regarding potential predictor variables, which need to be further examined in a larger sample using standardised measures of carer burden.

Keywords: Carer Burden, Stress, Predictors, Children, Caregivers, Parents,

Abbreviations

ZBI – Zarit Burden Interview
PSI – Parenting Stress Index
PIP – Paediatric Inventory for Parents

1. Introduction

Childhood chronic conditions are health conditions which last or are expected to last three months or longer, or those which are expected to reoccur (Mokkink et al., 2008). The prevalence of childhood chronic illness has been increasing and is estimated to continue to increase due to new medical interventions and treatments which decrease mortality rates (Liu, 2015). Childhood illness impacts both the child and the family; it has been linked to increased childhood mental health difficulties (Coburn et al., 2019) and difficulties with family functioning (Cousino & Hazen, 2013). Particularly, primary caregivers face the additional duties involved in caring for a sick child, such as treatment, medical appointments, role changes and the reality of changing expectations for their child's future (Smith et al., 2015). These challenges are unique to caregivers of children with chronic conditions and associated with increased mental health difficulties in caregivers (Pinquart, 2018). Above population norm levels of distress and mental health difficulties have been identified in caregivers of children with diabetes (Hessler et al., 2016), asthma (Kopel et al., 2017), epilepsy (Reilly et al., 2018), and cystic fibrosis (Brucefors et al., 2015) among others. In addition, caregivers of children with a chronic illness have been found to have higher rates of burnout (Lindstrom et al., 2010) as well as increased levels of depressive and anxiety symptoms (Van Oers et al., 2014) than caregivers whose child has no health condition.

The high incidence of mental health difficulties experienced by caregivers has been linked to the stress or burden experienced by caregivers of children with chronic conditions such as cancer (Santo et al., 2011). Caring for a child with a long term condition has been shown to result in significantly increased levels of stress in primary caregivers in comparison to caregivers of healthy children (Cousino & Hazen, 2013; Pinquart, 2018). The stress experienced by caregivers as a consequence of their caring role has been termed carer burden (also referred to as caregiver burden or stress, or parental burden or stress in literature). Carer burden has been defined as the physical, psychological, social and financial difficulties experienced by caregivers as a result of their caring role (Whalen & Buchholz, 2009). This increased stress or burden has also been associated with lower quality of life in caregivers of children with chronic conditions (Collins et al., 2020).

It is important to gain a better understanding of the factors which might predict carer burden in caregivers of children with chronic conditions in order to develop appropriate interventions and support for caregivers early on to curtail the impact on caregiver wellbeing. This population has not been extensively researched, and many caregivers feel that support has not been available to them throughout their caregiving role to assist them in dealing with the emotional impact of their child's condition (George et al., 2007). Caregivers have expressed a need for further support and information about ways to cope with their new caregiving role (Smith et al., 2015).

Research has demonstrated clear links between caregiver mental health difficulties and child outcomes including child wellbeing, management of the condition, and adjustment to the diagnosis (Palermo & Chambers, 2005; Whittemore et al., 2012). Addressing caregiver wellbeing in this population is therefore important, not only because of the impact of stress and mental health difficulties on caregivers but the impact of this on the child with a chronic condition. If the caregiver becomes unwell, this not only impacts on them but also influences their ability to care for their child (Lopez-Vargas et al., 2019). A number of research priorities have been identified by clinicians and caregivers of children with chronic conditions, and these included supporting caregivers and alleviating carer burden (Lopez et al., 2019). In order to better support caregiver's with the experience of burden, it is essential to be able to determine what factors might predict carer burden in caregivers of children with chronic conditions.

A review in 2013 began to collate findings regarding possible predictors of parenting stress in caregivers of children with chronic illnesses (Cousino et al., 2013). This review examined whether disease specific factors or parental cognitive factors (coping and appraisal) influence parenting stress. The review highlighted a number of potential predictors of carer burden including greater parental responsibility for treatment and management and lower levels of child self-care, and increased levels of child pain or frequency of painful episodes. The authors also highlighted a number of cognitive and appraisal factors which were related to increased burden including parental negative perceptions about the child's illness, feeling less able to manage the child's condition and viewing the child as vulnerable. The review found no link between burden and illness duration or severity. While this review (Cousino et al., 2013) was essential in beginning to detangle predictors of parenting stress in parents of children with long term conditions, it also examined a number of other factors including measures of parenting stress, levels of parenting stress and outcomes of parenting stress on child wellbeing. The focus of the results was therefore spread across a number of review questions and so may not have provided as much detail into specific predictors of parenting stress.

In addition the review also focused on specific illnesses (asthma, cancer, cystic fibrosis, diabetes, epilepsy, juvenile rheumatoid arthritis, and sickle cell disease). While these are some of the more common conditions experienced by children, the specific focus of this review may have resulted in some useful research in other disease populations being excluded based on the selection criteria. Additionally, the review search strategy only examined the concept of carer burden by using search terms such as parenting stress, family stress, and caregiver stress. The literature on carer burden has a number of synonymous terms for parenting stress including caregiver, carer and parental burden and as such this search strategy may have missed a number of studies examining burden. This is a difficulty in many studies examining carer burden in this population, and it's important to distinguish between generic stress and carer burden (Bastawrous, 2013).

A search of Cochrane Database of Systematic Reviews, Prospero, Google Scholar, PsychINFO and MEDLINE databases for similar reviews showed that no further reviews had been conducted to examine predictors of carer burden in caregivers of children with long term conditions following from Cousino and colleagues (2013). As the review by Cousino and colleagues (2013) examined papers published until mid 2012 an updated review of studies examining predictors of carer burden in this population is warranted. The present review updates the evidence base regarding predictors of carer burden in caregivers of children with chronic conditions.

As highlighted, the impact of caring for a child with a chronic condition has a detrimental impact on carer burden and mental health and as such it is important to consider what predictive factors may contribute to this in order to develop early identification, intervention and prevention. In order to address some of the limitations of the previous review, the current review focused solely on predictors of carer burden in order to be able to summarise more clearly what factors have been found to be associated with increased carer burden across health conditions and incorporated both the terms stress and burden in order to capture as many studies as possible.

2. Method

The proposal for this review was registered and accepted by Prospero on January 7th, 2020 with the ID number CRD42020161273. The search was carried out in March 2020.

2.1 Inclusion and exclusion criteria

Studies were included in the review if they a) were a quantitative and original research paper, b) focused primarily on carer burden or caregiver stress which were assessed using validated psychometric measures, c) had a sample of caregivers who look after children under the age of 18 with a physical long term health condition, d) were published between January 2012 and March 2020 e) were written in the English language f) had a full text available.

There were no limitations applied based on the type chronic conditions, so long as the condition lasts or is expected to last 12 months or longer. For instance medical conditions where the child was expected to recover relatively soon (such as an acute limb break) were excluded while chronic conditions such as severe allergies or heart conditions were included as these would likely impact their daily life.

Studies were excluded from the analysis where they a) were qualitative in nature, b) were a review article, book, commentary or similar c) were not accessible as a full text in English, d) the primary focus was on paid carers or professional/clinical staff, e) did not use a validated measure of carer burden or stress f) included caregivers of children over the age of 18. It was decided post-hoc that studies which focused on neurodevelopmental conditions, intellectual disabilities or mental health conditions would also be excluded, as these were a substantial portion of articles identified and this was beyond the scope of this review. As such studies which focused on neurodevelopmental or mental health conditions or had a mixed sample of conditions which was not easily distinguishable in the data were excluded.

2.2 Search Strategy

An expert librarian was consulted in the development of the search strategy. The primary strategy involved searching the following electronic databases: PsychINFO, MEDLINE, EMBASE, Cumulative Index to Nursing and Allied Health Literature. Pre-determined search terms were used across three stem groups in the order below:

1. The first stem group related to carer burden and included the search terms (Caregiver burden OR parental burden OR carer burden OR care* stress OR parent stress OR family stress OR maternal stress OR maternal burden OR paternal stress OR paternal burden).
2. The second stem group related to children and included the search terms (child* OR pediatric OR paediatric OR young person OR teen* OR adolescent OR baby OR infant OR youth).
3. The third search stem related to long term conditions and included the search terms (chronic illness OR long-term condition OR illness OR disease OR disability OR health cond* OR neurodevelopmental disorder OR developmental disabilities OR learning disabilities).
4. Results from the searches for each of the three stems were combined with AND. These were then combined with results from a further stem related to predictors which used the search terms (Predict* OR risk* OR risk factors).

The search results were then limited to articles published between January 2012 and March 2020, English language and full text availability. Results were de-duplicated. The secondary search strategy involved hand searching relevant articles from reference lists. A grey literature search using Google Scholar was also carried out which examined the first 200 results, and the reference lists of included articles from databases were reviewed to identify further relevant studies.

2.3 Quality Assessment and Risk of Bias

The methodological quality of studies was assessed using an adapted version of the Agency for Healthcare Research and Quality (AHRQ) risk of bias tool. This tool was recommended for cross sectional studies by Zeng and colleagues (2015). The adapted version excluded questions based on follow up and control groups as this was not deemed relevant for the included studies. The adapted tool assesses risk of bias based on cohort selection and inclusion criteria, sample size and description, use of validated measures and appropriateness of analysis including confounding factors. The AHRQ is rated using a 3 point Likert scale (0 = No, 1 = partial, 2 = yes or N/A if this is not applicable). The maximum possible score for the adapted version was 16. Risk of bias assessment was completed by the primary researcher for all studies, and two independent individuals double rated a total of 58% of studies.

2.4. Data Extraction and Synthesis

Data was extracted using a pro-forma including details on: a) year of publication, b) location, c) authors, d) child health conditions, e) study design, f) sample characteristics, g) inclusion/exclusion criteria, h) measures used (carer burden and other variables, i) relevant findings, and j) summary of predictors measured. The data extraction form can be found in Table A1 in Appendix A. It was beyond the scope of this review to examine specific predictors of subscales of carer burden measures, and as such only predictors of total scores were examined unless only subscale data was presented. Data was extracted by the primary researcher, and a second rater independently extracted 30% of data to ensure that the primary researcher was not influenced by bias. A high level of agreement was found between the independent rater and the primary researcher in terms of which relevant information was extracted from the studies, and any differences were discussed and amended appropriately.

Data was examined using a narrative synthesis approach, which has been recommended for use in reviews where a variety of measures and populations are examined (Popay et al., 2006). This approach was selected during the planning phase due to the nature of studies, varied measures of carer burden and the need for preliminary synthesis of findings. Firstly the researcher summarised the findings from each study, as well as pertinent characteristics about each study by extracting this data into an Excel spreadsheet. Study findings were then grouped by various factors (including type of condition, type of analysis, sample selection, measures used and quality rating) in order to examine potential patterns in findings. A vote counter approach was used to provide an initial description of findings, and data was then transformed into a descriptive account of findings and patterns across studies taking into account quality assessment and other potential variations within and between studies.

3. Results

3.1 Search Results

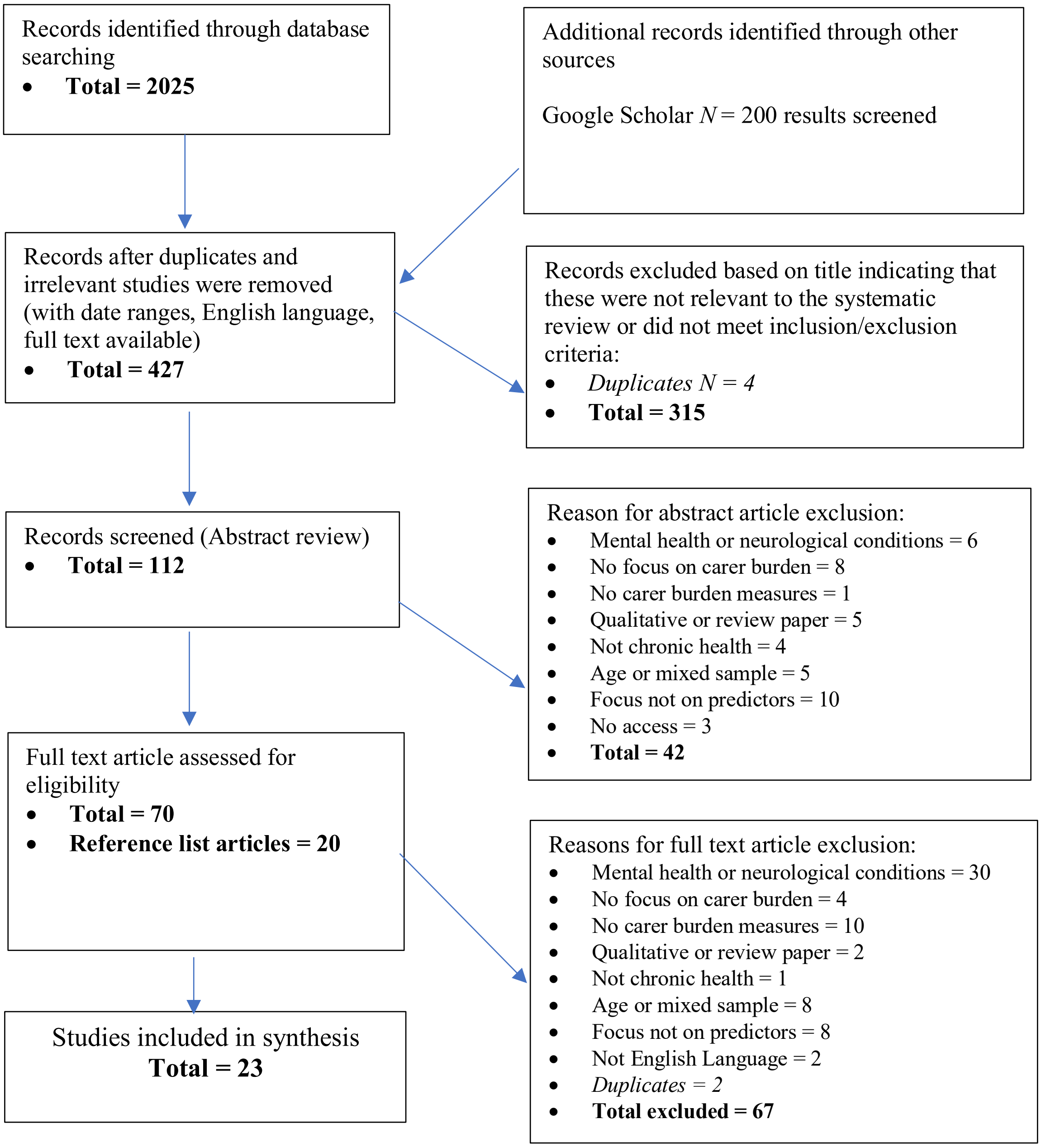
As illustrated in Figure 1 below 2225 papers were originally found following the search via databases and a grey literature search using Google Scholar. The author read through the titles of all articles identified, excluding any which were not deemed relevant to the current review. Articles which seemed relevant were then examined by reading the abstract, and any abstracts which clearly showed that inclusion criteria were not met were excluded. Following this, articles which met inclusion and exclusion criteria were read in full. From this set of articles, the final included articles for the review were selected. For details regarding reasons for exclusion see Figure 1 below.

3.2 Included Studies

A total of 23 studies were included in this review.

Figure 1.

PRISMA Flow Chart of Included Studies



Records identified through database searching
- **Total = 2025**

Additional records identified through other sources

Google Scholar *N* = 200 results screened

Records after duplicates and irrelevant studies were removed (with date ranges, English language, full text available)
- **Total = 427**

Records excluded based on title indicating that these were not relevant to the systematic review or did not meet inclusion/exclusion criteria:
- *Duplicates N = 4*
- **Total = 315**

Records screened (Abstract review)
- **Total = 112**

Reason for abstract article exclusion:
- Mental health or neurological conditions = 6
- No focus on carer burden = 8
- No carer burden measures = 1
- Qualitative or review paper = 5
- Not chronic health = 4
- Age or mixed sample = 5
- Focus not on predictors = 10
- No access = 3
- **Total = 42**

Full text article assessed for eligibility
- **Total = 70**
- **Reference list articles = 20**

Reasons for full text article exclusion:
- Mental health or neurological conditions = 30
- No focus on carer burden = 4
- No carer burden measures = 10
- Qualitative or review paper = 2
- Not chronic health = 1
- Age or mixed sample = 8
- Focus not on predictors = 8
- Not English Language = 2
- *Duplicates = 2*
- **Total excluded = 67**

Studies included in synthesis
Total = 23

3.2.1 Study characteristics

The characteristics of the included studies and demographic details of participants are summarised below in Table 1. The majority of studies (*N* = 22) were cross sectional, with one having a longitudinal design (19). Most participants were recruited from routine medical appointments, only three studies recruited online (3, 6, 18). The largest proportion of studies focused on caregivers of children with cancer (*N*=6), were completed in the United States (*N*=7) and were published in 2016 (*N*=4). The total sample size of all studies was 3555 and sample sizes ranged from 49 to 495 with a mean sample size of 155 participants. The mean age of caregivers across studies was 37.5 years (mean age range 30 years old to 45.7 years old). Five studies did not report caregiver age (4, 9, 12, 17, 22). Participant samples were predominantly female with a mean across studies of 76.8% female participants (range 50% to 100%). Two studies used a sample of 100% female participants (2, 19) and three studies did not report the gender of participants (5, 9).

The included studies used a wide range of measures for carer burden. The majority of studies used the Pediatric Inventory for Parents (*N*=7) followed by the Zarit Burden Interview (*N*=6). A summary and description of the measures used can be found in Table B1 in Appendix B.

Table 1

Summary of Study Characteristics

Study Location Condition	**Study Design**	**Sample Characteristics**	**Child Characteristics**	**Inclusion/Exclusion Criteria**
Asadullah et al. 2017 India Human Immunodeficiency Viruses	Cross sectional	***N*** = **171** caregivers recruited from appointments **Demographic data** Gender: Female 64.9% Age: M 39.2 years (SD 10.9)	Gender: Female 50.3 % Age: M 10.8 years (SD 3.9)	Caregivers of children infected with Human Immunodeficiency Viruses.
Boztepe et al. 2019 Turkey Leukaemia and cerebral palsy	Cross sectional	***N*** = **69** mothers of children with cerebral palsy recruited from a research hospital (control group) **Demographic data cerebral palsy** Gender: Female 100% Age: M 34.6 years (SD 7.3) range 21-54 ***N*** = **70** mothers of children with leukaemia recruited from a haematology outpatient service. **Demographic data Leukaemia** Gender: Female 100% Age: M 35.5 years (SD 6.7)	**Cerebral palsy:** Age: M 7.4 years (SD 4.0) Gender: Male 58% **Leukemia:** Age: M 7.1years (SD 3.7) Gender: Male 55.8%	Caregivers of children under 18 with leukaemia or cerebral palsy (control), with no current mental illness. Children with leukaemia must be at least 6 months past diagnosis, have no additional physical illness, and be in active treatment.

Caris et al. 2016 Online Heart Disease	Cross sectional	***N* = 459** caregivers recruited online **Demographic data** Gender: Female 86.9% Age: M 35.3 years (SD 7.2)	Gender: Male 63% Age: M 4.9 years (SD 5.2)	Caregivers of children with hypoplastic left heart syndrome
Chivukula et al. 2018 India Acute lymphoblastic leukaemia	Cross sectional	***N* = 100** caregivers recruited from hospitals **Demographic data** Gender: Female 50%, Male 50%	Age range: 3-11 years	Caregivers children between the ages of 3–11 years diagnosed with leukaemia. Excluded caregivers of children in palliative care or those who had external care for their children.
Crespo et al. 2016 Portugal Cancer	Cross sectional	***N* = 204** caregivers recruited from hospitals **Demographic data** Relationship to child: Mother 87.3%, father 10.8%, other relatives 2% Age: M 40.11 years (SD 7.12)	Age: M 11.19 years (SD 4.98) Diagnosis: • Leukemia: 43.1% • Solid tumors: 22.5% • Lymphomas: 17.6% • Central nervous system: 15.2% • Langerhans cell histiocytosis: 1.5%	Primary informal caregivers for a child with cancer at least 3 months post diagnosis
Dekoven et al. 2014 United States of America Haemophilia	Cross sectional – online survey	***N* = 310** caregivers recruited online **Demographic data** Gender: Female 89.4%, Male 10.7% Age: • 18 – 34: 24.5% • 35 – 44: 47.7% • 45 – 54: 26.5% • 55 – 64: 1.3%	Diagnosis: in the last 6 months 3.2%, 6 – 12 months ago 5.2%, 1 – 2 years ago 5.8%, more than 2 years ago 85.8% Type of diagnosis: Haemophilia A 86.5%, B 10.7% and other 1.6% Severity: Mild 5.8%, moderate 10.7%, severe 83.6%	Caregiver over the age of 18, caring for a child with haemophilia, fluent in English.

Edmond et al. 2016 United States of America Cancer (32.3%) Sickle cell disease (51%) Haematological conditions (16.7%)	Cross sectional	***N* = 96** primary caregivers recruited through outpatient visits at the clinic. **Demographic data Sickle cell disease:** Gender: Female 85%, Mothers 79% Age: M 37 years (SD 8) **Demographic data Cancer:** Gender: Female 90.3%, Mothers 83.9% Age: M 37.1 years (SD 9) **Demographic data other conditions:** Gender: Female 93.8%, Mothers 87.5% Age: M 30.4 years(SD 7.9)	**Sickle cell disease:** Age: M 10.8 years (SD 4.5) Gender: 49% female **Cancer:** Age: M 11.2 years (SD 5) Gender: Female 54.8% **Other conditions:** Age: M 7.9 years (SD 4.8) Gender: Female 25%	Caregivers of children under the age of 18 with cancer, sickle cell disease and other haematological conditions. English speaking caregivers without developmental delays or cognitive impairment.
Fitzgerald et al. 2018 Ireland Cystic Fibrosis	Cross sectional	***N* = 130** mother & father dyads recruited as part of a larger study**.** **Demographic data** Gender: Female 50 % Age: Mothers M 35.5 years (SD 4.9), Fathers M 38 years (SD 5.4)	Age: M 37.6/37.3 months (range 17.4 – 61.9/4).	Caregivers of children with cystic fibrosis without another child in the main study.
Gray et al. 2013 United States of America Pediatric Inflammatory Bowel Diseases	Correlational – validating a psychometric measure	***N* = 130** adolescents recruited from a larger study. **Demographic data** Relationship status: 80% Married, 80% mothers of the children; 12.4% Father, 7.7% Unknown	Gender: 61% female Age: 15.64 +/- 1.36	Caregivers of adolescents (13-17 years) with a diagnosis of Crohn's disease or ulcerative colitis. Fluent in English Excluded patients with corticosteroid greater than 1 mg/kg/day. No other chronic illness in the adolescent. No neurocognitive disorder.

Guilfoyle et al. 2012 United States of America Inflammatory Bowel Disease	Cross sectional	***N* = 65** parents of children with inflammatory bowel disease recruited from a larger trial. **Demographic data** Gender: 90.3% female Age: M 45.7 (SD 5.6)	**Child characteristics:** Age: M 15.5 (SD 1.4) Gender: 43% female	Primary parents of a child aged between 13 and 17 with a diagnosis of Crohn's disease or ulcerative colitis. Excluded if they had a neurocognitive disorder, comorbid chronic medical condition or were not fluent in English.
Jubber et al. 2013 United States of America Type 1 diabetes	Cross sectional	***N* = 85** children with Type 1 diabetes and their parents recruited from clinics, summer camps and referrals. **Demographic data** Gender: 93%of families had two parents Age: Mothers M 39.46 (SD 5.93) Fathers M 42.37 (SD 5.9)	Age: M 12.6 (SD 3.26) Time since diagnosis: M 4.69 (SD 3.41)	Child with Type 1 diabetes, sibling between 8 and 18 years old and willing to participate.
Kaugars et al. 2018 United States of America Congenital Heart Disease	Cross sectional	***N* = 54** parents recruited from assessment by a psychologist. **Demographic data** Relationship status: married 61.11%	Gender: Male 85.19% Age: M 7.48 years (SD 2.38)	Caregivers of a child aged 3 – 13 with congenital heart disease seen by the psychologist Excluded extremely low defensive responding scale scores
Kobos et al. 2015 Poland Diabetes	Cross sectional	***N* = 112** direct caregivers recruited from diabetes clinics. **Demographic data** Gender: 50% fathers 50% mothers Age: M 39.6 years (SD 6.8) mothers with 34% under 35 years old M 43.2 years (SD 7.6) fathers with 34.8% above 45 years old.	Age: M 11.3 years (SD 3.6), 49.1% primary school age, 34.8% middle school age, 16.1% nursery age. Gender: 52.7% female, 47.3% male	Diagnosis of Type 1 diabetes for the child for 6 months plus. Child aged between 3 and 16 years. Siblings without a chronic illness. Direct caregivers who live with the child and spend the largest amount of time with the child in comparison to others.

Olagunju et al. 2015 Nigeria Obstructive Adenotosillar hypertrophy	Cross sectional	***N* = 100** caregivers recruited from health centres **Demographic data** Gender: Female 84% Age: M 34.3 years (SD 5.4)	Age: M 36.7 months (SD 21.8) Gender: Male 68%	A diagnosis of Obstructive Adenotosillar hypertrophy for the child. Caregivers directly involved in the care of the child, not a primary caregiver for anyone else at this time.
Piran et al. 2016 Iran Chronic disease	Cross sectional	***N* = 249** caregivers recruited via hospital or ambulatory service referrals **Demographic data** Gender: 89.6% female Age: M 35.4 (SD 7.6)	Age: M 7.27 years (SD 4.06) Gender: 53.8% female	Caregivers over 18 providing at least 3 months of care to a sick child (main caregiver) Children: 14 years or younger with at least one chronic disease diagnosed. Excluded caregivers with a psychiatric disorder.
Rubira et al. 2012 Brazil Cancer	Cross sectional	***N* = 160** caregivers recruited from an oncology service via random sampling. **Demographic data** Gender: 88.75% female, 11.25% male Age: M 34.96 years, range 18 - 57	Age: M 9.09 (range 1 – 17) Gender: 55% male, 45% female	Caregivers of children with a diagnosis of cancer who were undergoing chemotherapy for a minimum of two months. Aged 18 or over, provide care for the patient for majority of the time, live with the patient.
Sarajuuri et al. 2012 Finland Univentricular heart defects	Cross sectional	***N* = 23** patients with hypoplastic left heart syndrome and **14** with Univentricular heart recruited as part of a prospective neurodevelopmental follow up study and **46** healthy gender matched new-borns as controls. **Demographic data**	**Hypoplastic left heart syndrome :** Age: M 18 months (range 17-20) Gender: 79% male **Univentricular heart:** Age: M 18 months (range 18-20) Gender: 46% male	Caregivers of patients with hypoplastic left heart syndrome or Univentricular heart born between 2002 and 2005 operated on in the Helsinki University Central Hospital. No

		Occupation level: **Hypoplastic left heart syndrome :** M 4 (professionals) **Univentricular heart :** M 3 (technicians & assoc. professionals) **Control:** M 3 (technicians & assoc. professionals)	**Control:** Age: M 18 months (range 18-23) Gender: 65% male	known chromosomal abnormalities in children. Child aged 18 months.
Senger et al. 2016 United States of America Mitochondrial disease	Cross sectional	***N* = 231** parents of children with mitochondrial disease recruited online. **Demographic data** Age: M 42.3 years (SD 8.3) Gender: 95% Mothers	Age: M 9.85 years (SD 5.24) Gender: female 51%, male 49%	Parents of at least one child with a confirmed, probable, or possible diagnosis of mitochondrial disease.
Sulkers et al. 2015 Netherlands Cancer	Longitudinal	***N* = 95** mothers of children with cancer recruited from a medical centre. **Demographic data** Gender: 100% female sample Age: Range 24-55	Age: M 8.4 years (SD 5.3) range 0-17 Gender: Female 54%	Primary caregiver of a child with cancer aged 0 to 18 years with no prior diagnosis of cancer, receiving treatment. Fluent in Dutch.
Toledano – Toledano et al. 2019 Mexico Chronic conditions	Cross sectional	***N* = 416** recruited from a hospital. **Demographic data** Gender: 81.7% female, 18.3% male Age: mothers M 31.05 years (SD 7.74) fathers M 34.43 years (SD 8.71)	Age: M 5.91 years (SD 5.03) Gender: 47.4% female, 52.6% male	Parents aged 18 and over who were the caregiver of a child with a chronic disease and required specialised hospital treatment. Sufficient literacy skills.
Torres – Ortuno et al. 2014 Spain Haemophilia	Cross sectional	***N* = 49** parents of children with haemophilia recruited during a workshop **Demographic data** Gender: Female 53.1%, male 46.9% Age: 38.86 (SD 3.26)	Age: 4.02 years (SD 2.01)	Attending a workshop for parents of children with haemophilia.

Wang et al. 2017 China Acute lymphoblastic leukemia	Cross sectional	***N* = 130** primary caregivers of children diagnosed with Acute lymphoblastic leukemia – secondary data **Demographic data** Gender: 26.9% male, 73.1% female	Age: • Under 1: 6.1% • 1-3 years: 29.2% • 3-7 years: 33.1% • 7-12 years: 23.1% • 12-18 years: 8.5% Gender: 63.8% male, 36.2% female	Parents who had a child under 15 years old with a new diagnosis of Acute lymphoblastic leukemia. High school diploma or higher education and fluent Mandarin Chinese. Monthly family income above 2000 Yuan.
Zhu et al. 2019 China Pediatric Glaucoma	Cross sectional	***N* = 57** recruited from an ophthalmic centre **Demographic data** Gender: 33% male, 66.7% female Age: M 30.02 (SD 4.85)	Age: M 30.09 months (SD 35.11) Gender: 66.7% male, 33.3% female	Parents of children with pediatric glaucoma
Note: Abbreviations		*SD = Standard deviation, M = mean*		

3.2.2 Quality Assessment and Risk of Bias

Overall Quality. The quality assessments per study can be found in Table 2. The inter-rater agreement was calculated by comparing the number of concurrent ratings between the primary and secondary raters, and was found to be 96.4%. Where study ratings differed, scores were discussed until these were mutually agreed upon. Scores on the AHRQ ranged from 37.5% to 87.5% with a mean rating of 71.5%. The overall study quality seemed acceptable, with some variation in scores on the AHRQ. However, there is little merit of examining quality based on numerical scores and it is more helpful to identify patterns of quality or risk of bias issues across the literature in order to support future research as suggested by Juni and colleagues (1999). There was only one study which appeared to be different from the majority in terms of quality (Torres-Ortuno et al., 2014). This study did not report sample size calculations, inclusion or exclusion criteria, have an adequate description of the cohort or control for confounding variables. A further study (Sarajuuri et al., 2012) did not include demographic information about their sample in the paper and it was difficult to track further information in previous publications of the same data. It is recommended that all pertinent data be presented in each publication or within an Appendix.

Table 2

Quality Assessment of included studies using AHRQ Score 2=Yes Score 1 = Partially Yes Score 0 = No Can't tell or N/A

Study	Unbiased selection of the cohort	Sample size calculated/ 5% difference?	Adequate description of the cohort?	Inclusion and exclusion criteria clear?	Validated Carer Burden measure?	Validated measures of predictors used?	Analysis controls for confounding ?	Analytic methods appropriate?	**Total Score (%)**
1 Asadullah et al., 2017*	1	2	2	1*	2	1	0	2	11/16 (68.0)
2 Boztepe et al., 2019	2	0	2	2	2	1	1	2	13/16 (81.3)
3 Caris et al., 2016	1	0	2	2	2	2	0	2	11/16 (68.8)
4 Chivukula et al., 2018*	1*	0	1	2	2	2	0	2	10/16 (62.5)
5 Crespo et al., 2016	1	0	1	2	2	2	1	2	11/16 (68.8)
6 Dekoven et al., 2014	1	0	2	2	2	1	2	2	12/16 (75.0)

Study	Unbiased selection of the cohort	Sample size calculated/ 5% difference?	Adequate description of the cohort?	Inclusion and exclusion criteria clear?	Validated Carer Burden measure?	Validated measures of predictors used?	Analysis controls for confounding ?	Analytic methods appropriate?	**Total Score (%)**
7 Edmond et al., 2016*	1	2	2	2	2	2	1	2*	14/16 (87.5)
8 Fitzerald et al., 2018	1	0	2	2	2	1	1	2	11/16 (68.8)
9 Gray et al., 2013	2	0	0	2	2	2	0	2	10/16 (62.5)
10 Guilfoyle et al. 2014	1	0	2	2	2	2	2	2	13/16 (81.3)
11 Jubber et al., 2013*	2	1	2	1	2*	2	2	2	14/16 (87.5)
12 Kaugars et al., 2018*	1*	0	1	1	2	2	0	2	09/16 (56.3)
13 Kobos et al., 2015*	1	0	2*	2	2	0	2	2	11/16 (68.8)
14 Olagunju et al., 2015	1	0	1	1	2	2	0	2	09/16 (56.3)
15 Piran et al., 2017	1	1	2	2	2	1	1	2	12/16 (75.0)
16 Rubira et al., 2012	1	0	2	2	2	2	1	2	12/16 (75.0)

Study	Unbiased selection of the cohort	Sample size calculated/ 5% difference?	Adequate description of the cohort?	Inclusion and exclusion criteria clear?	Validated Carer Burden or parenting stress measure?	Validated measures of outcome used?	Analysis controls for confounding ?	Analytic methods appropriate?	**Total Score (%)**
17 Sarajuuri et al., 2012	1	2	0	2	2	1	2	2	12/16 (75.0)
18 Senger et al., 2016	1	0	2	2	2	2	1	2	12/16 (75.0)
19 Sulkers et al., 2015	2	0	2	2	2	1	1	2	12/16 (75.0)
20 Toledano-Toledano et al., 2019	1	0	2	2	2	2	2	2	13/16 (81.3)
21 Torres-Ortuno et al., 2014	1	0	0	0	2	1	0	2	06/16 (37.5)
22 Wang et al., 2017	2	0	2	2	2	2	1	2	13/16 (81.3)
23 Zhu et al., 2019*	2*	0	2	2	2	2	0	2	12/16 (75.0)

Note: * denotes a study or point where the second rater disagreed, and these disagreements were discussed and resolved.

Selection, sample size, and description of the cohort. The majority of studies (*N*=15) were rated as having a partially unbiased sample selection process as they recruited their participants from clinics or hospitals based on the child's health condition (1, 2, 4, 5, 7, 11, 12, 13, 14, 15, 16, 19, 20, 22, 23). Three studies recruited participants online (3, 6, 18), four studies recruited from a larger study sample (8, 9, 10, 17), and one study recruited from a workshop for caregivers (21). These studies offered the opportunity to participate to all eligible participants visiting the hospital, while a few advertised for volunteers. This was deemed an appropriate method of sample selection given the need for the child to have a particular health diagnosis.

Sample size calculations were not often reported. Only three studies fully described their sample size calculations (1, 7, 17) and a further two partially described their sample size estimates (11, 15). While most samples appeared to have sufficiently large samples, it is imperative that research adequately describes the sample size and how this affects the effect sizes found even in non-RCT studies.

Most studies were deemed to have an adequate (*N* =16) or partially adequate (*N* = 4) description of the cohort. Three studies did not have an adequate description of their sample, often missing key demographic data about caregivers while describing the children in more detail (9, 17, 21). Reporting key information such as caregiver gender and age while investigating carer burden seems crucial, yet three studies failed to report age (4, 5, 9) and five failed to report gender (4, 9, 12, 17, 22).

Inclusion criteria and measures used. All studies presented clear inclusion and exclusion criteria with the exception of one study which was rated as non-adequate as it described the inclusion criteria as attending a group (21) and was unclear whether further criteria were applied. All studies used a validated carer burden or parenting stress measure, as this was an inclusion criterion for this review. Most studies used additional measures or demographic questionnaires to examine possible predictors of carer burden or parenting stress, and these were deemed to be valid for the construct being measured.

Data analysis. All studies were deemed to have an appropriate analytic method, with most ($N = 19$) utilising correlational or regression methods (1, 2, 3, 4, 5, 7, 9, 10, 12, 13, 14, 15, 16, 17, 18, 20, 21, 22, 23). Two studies used general linear models (6, 8) and two studies used multi-level and structural equation modelling (11, 19). However eight studies did not state that they controlled for confounding variables (1, 3, 4, 9, 12, 14, 21, 23). Future studies should ensure that they control for confounding variables and explicitly state this.

3.2.3 Key Study Findings

The key findings of the included studies and measures used can be found in Table 3 below.

Table 3

Summary of Key Findings

Study	**Carer burden/parental stress measures**	**Other measures**	**Predictors of higher burden identified**	**Predictors of lower burden identified**	**No evidence or risk found**
Asadullah et al. 2017	Zarit Burden Interview	General Health Questionnaire-12	• Biological parents		• Smoking status • Alcohol consumption • Caregiver gender • Schooling • Family type • Religion
Boztepe et al. 2019	Zarit Burden Interview	Beck Depression Inventory Severity of illness scale devised by researchers	• Higher illness severity • Depression • Maternal age • Having a male child • Lower child age		
Caris et al., 2016	Paediatric Inventory for Parents	Parenting Stress Index Short Form Pediatric Quality of Life Inventory	• Lower caregiver age • Lower child age • Other medical issues • Neurodevelopmental issues		• Caregiver gender • Ethnicity • Education • Relationship status • Living location
Chivukula et al. 2018	The Caregiver Burden Inventory	The Brief Cope The Spirituality Scale	Emotional burden: • Self-distraction • Denial • Substance use • Use of emotional and instrumental support • Behavioural disengagement	Developmental burden: • Positive reframing • Acceptance • Religion • Self-discovery • Relationships • Eco-awareness	• Caregiver gender

			• Self-blame • Venting • Positive reframing • Acceptance • Relationships • Eco-awareness Physical burden: • Self-distraction • Denial • Behavioural disengagement • Self-blame Social burden: • Behavioural disengagement	Physical burden: • Use of emotional support • Positive reframing • Planning • Acceptance • Self-discovery • Relationships Social burden: • Use of emotional support and instrumental support • Positive reframing • Planning • Acceptance • Religion • Self-discovery • Relationships • Eco-awareness.	
Crespo et al. 2016	Revised Burden Scale – objective burden subscale (Portuguese version)	Care-20 EUROHIS-QOL-8 Satisfaction with Life Scale		• Higher child age • Higher caregiver age • More time since diagnosis • Experiencing family centered care	
Dekoven et al. 2014	Carer QoL	Novel burden questionnaire Demographic questionnaire	• Current presence of an inhibitor • Higher number of hours of work lost due to illness • Two or more bleeding episodes experienced by child in a month	• Higher income	• Caregiver gender • Caregiver age • Education • Marital status • Family income. • Disease severity or hemophilia type

					Distance from the center Time since diagnosis Treatment type.
Edmond et al. 2016	Impact on Family Scale	Pediatric Sleep Questionnaire Pain Questionnaire Child Behaviour Checklist Family Symptom Inventory	Financial difficulties Caring for a child with cancer Higher child pain Child behavioural difficulties	Employment	Caregiver age Caregiver gender Caregiver ethnicity Caregiver relationship status Caregiver education, Child age Child gender Child ethnicity Child sleep problems.
Fitzgerald et al. 2018	Carer QoL	Demographic questionnaire	Mothers (with child under40 months) Higher child age Being a mother Having a child test positive for pseudomonas aeruginosa.		Caregiver relationship status Diagnosis method Having meconium ileus Having other siblings with Cystic Fibrosis Mother’s education level
Gray et al. 2013	The Paediatric Inventory for Parents	The McMaster Family Assessment Device Pediatric Crohn's Disease Activity Index Lichtiger Colitis Activity Index Child Behavior Checklist and Youth Self-Report Demographic questionnaire	Poorer family functioning Adolescents reporting internalizing symptoms Disease severity for patients with Crohn’s disease		

Guilfoyle et al. 2012	Paediatric Inventory for Parents	Children's Depression Inventory Disease severity score using Pediatric Crohn's Disease Activity Index or Lichtiger Colitis Activity Index Demographic questionnaire	• Disease severity		• Child age • Child gender • Child ethnicity • Family income.
Jubber et al. 2013	Caregiver Strain Index	Porter O'Leary Scale (marital conflict) HbA1c from medical records Demographic information	• Mothers • Worse metabolic control (higher burden for fathers) • Increased marital conflict		• Caregiver years of education • Caregiver income
Kaugars et al. 2018	Paediatric Inventory for Parents	The Parenting Stress Index Short Form The Parenting Stress Index Short Form 4th Edition PedsQL Functional Independence Measure Demographics and illness-related characteristics	• Higher disease severity • Other medical conditions • Younger gestational age • Attention and behavioural difficulties		
Kobos et al. 2015	Caregiver Burden Scale	Interview	• Unemployment	• Higher child age	• Multigenerational households • Health conditions of parents • Method of insulin administration • Child gender

					• Family factors (place of residence, financial situation, number of children in the household, parent's age or education) • Clinical factors (duration of the disease, number of tests during the day or night, number of hospitalisations, metabolic control or frequency of episodes).
Olagunju et al. 2015	Zarit Burden Interview	General Health Questionnaire – 12 Demographic questionnaire	• Emotional distress		• Caregiver gender • Caregiver education • Child age • Child gender • Child academic delay • Child schooling
Piran et al. 2017	Caregiver Burden Scale	Demographic questionnaire	• Child with cerebral palsy > cancer > epilepsy • Higher level of disability and dependence • Caregiving to another family member	• Higher income • Larger family size	• Child age • Disease duration • Caregiver age • Caregiver education • Duration of care • Caregiver gender • Child gender • Caregiver employment status • Residency location • Caregiver health conditions
Rubira et al. 2012	Zarit Burden Interview	Short Form 36	• Receiving poor or very poor support from a partner		

		Demographic questionnaire	• Low literacy level or only elementary education • Higher number of children • Not receiving help to care for the child • Caregiver health problems • Being unemployed or a home maker • Black ethnicity.		
Sarajuuri et al. 2012	Parenting Stress Index	Achenbach Child Behaviour Checklist Demographic and clinical data	• Mothers experienced more parent related stress • Maternal occupation related to maternal stress • More burden for Hypoplastic left heart syndrome compared to control.		• Maternal skill level.
Senger et al. 2016	Paediatric Inventory for Parents	Coping Health Inventory for Parents Demographic questionnaire	• Higher hospital visits in the past year • Greater number of specialties involved • Higher number of organs involved • Higher number of medical office visits	• Higher caregiver age • Higher child age • Higher income • Higher family integration, social support and total Coping Health Inventory for Parents scores	
Sulkers et al. 2015	Paediatric Inventory for Parents – difficulty subscale	Intensity of Treatment Rating Scale Centre for Epidemiologic Studies Depression Scale State Trait Anxiety Inventory 4 items of the Health Perception Scale of the Short-	• Single mother • Having only one child		• Child gender or age • Caregiver age • Marital status • Number of children • Employment or education • Type of diagnosis • Prognosis • Treatment status

		Form General Health Survey (SF-20)			• Relapse or treatment severity
Toledano – Toledano et al. 2019	Zarit Burden Interview	Sociodemographic Variables Questionnaire (Q-SV) Family Support Questionnaire Social Support Network Scale Parental Stress Scale Family Functioning Scale WHO Wellbeing Index Historic-Psycho-Socio-Cultural-Premises Scale	• Following historical socio-cultural premises • Stressors • Anxiety.	• Higher education level • Better social support networks • Family support • Positive family functioning • Overall well-being.	• Caregiver gender
Torres-Ortuno et al. 2014	Paediatric Inventory for Parents	Faces III (family functioning) Data on characteristics & treatment	• Mothers experienced more frequent stress		• Severity of disease • Family history • Number of other children • Using an inhibitor or port-a-cath • Having a history of haem-arthrosis
Wang et al. 2017	Zarit Burden Interview	Perceived Social Support Scale Self-rating anxiety scale (Zung) Self-rating depression scale (Zung) Short Form -36 Demographic questionnaire	• Anxiety and depression • Daily care time	• Social support • Better general health • Higher average monthly family income • Social support • Number of co-caregivers	• Caregiver age or gender • Caregiver nationality, • Caregiver education • Marital status • Chronic disease • Religion • Profession or employment • Living location or distance from hospital

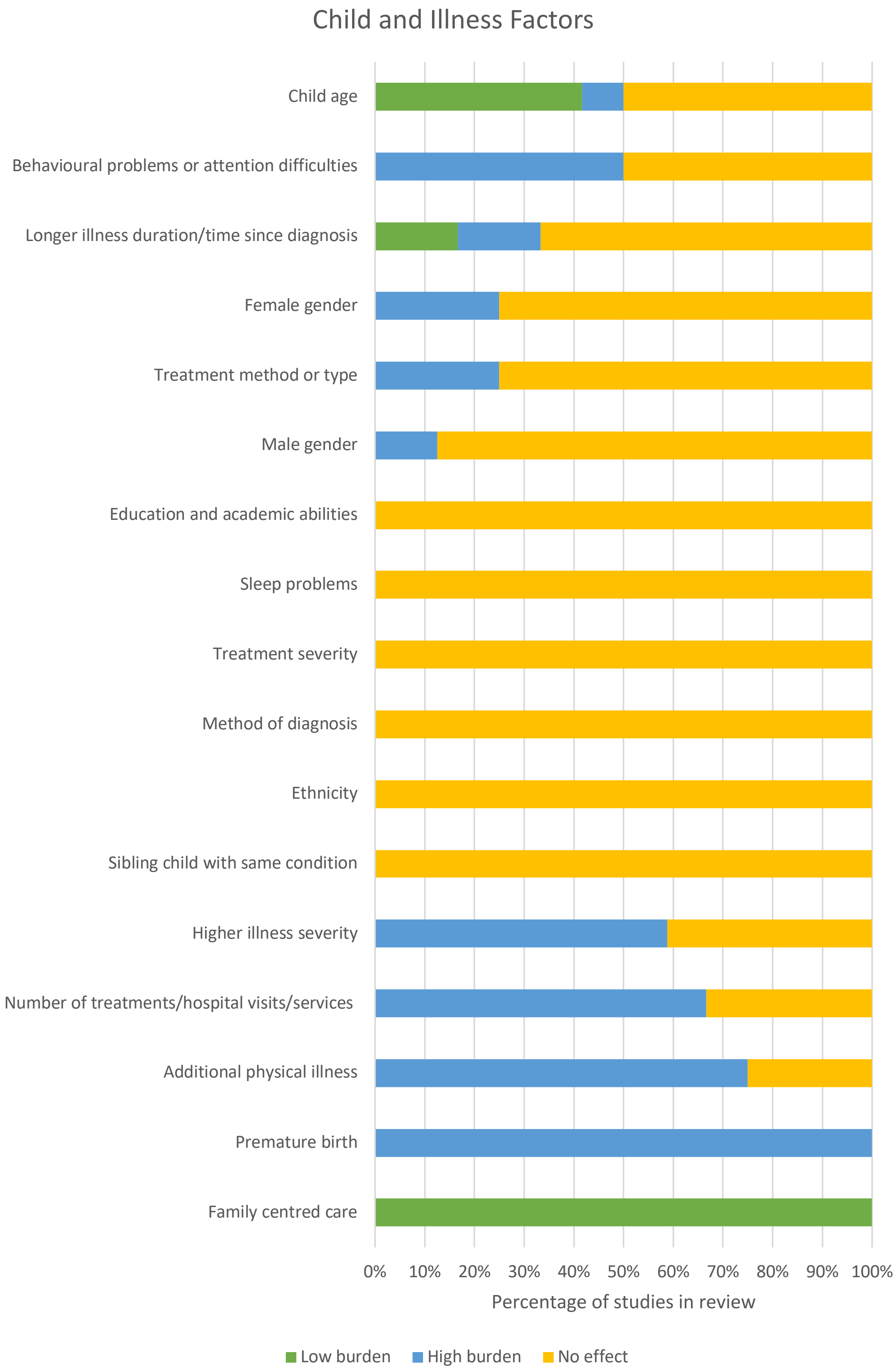
				• Child age • Child gender • Child education
Zhu et al. 2019	Caregiver Burden Inventory Positive Aspects of Caregiving Questionnaire	Demographic questionnaire	• Longer disease duration • Lower education level • Being a female caregiver • Being employed • Having a lower income	

3.2.4 Predictors of Carer Burden and Parenting Stress

The included studies examined a number of possible predictors of carer burden and parenting stress, including specific demographic characteristics of both the caregiver and the child as well as other validated outcome measures; 29 caregiver characteristics and 16 child or condition characteristics were examined across the studies.

Child characteristics and Illness related factors. A summary child characteristics and illness related factors found to predict carer burden in caregivers is presented below. Figure 2 below is a summary of these findings.

Figure 2. Graph displaying the percentage of studies showing evidence for links with lower, higher burden or no effect and child and illness factors.



Age. Twelve studies in total examined child age as a possible predictor. Six of the twelve studies (50%) suggested no effect of child age on caregiver burden (7, 10, 14, 15, 19, 22). Five of the twelve studies (41.6%) showed some effect, of which three studies suggested that older child age predicted lower burden (3, 5, 18) and three studies suggested that having a younger child was correlated with increased burden (2, 3, 13). Only one study (8) suggested that having an older child was associated with higher levels of burden for caregivers than having a younger child, and this study examined caregivers of children with cystic fibrosis. As this is a life limiting condition the increased burden may reflect that increased child age coincides with increased disease severity and caring needs. Caregivers of younger children may experience more burden due to normal developmental differences in younger children such as lower levels of communication and self-care skills. Additionally one study examined premature birth (12) and reported links with increased levels of burden.

Gender. Eight studies examined child gender as a possible predictor of carer burden. Seven of the eight studies (87.5%) suggested no link between gender and burden (7, 13, 19, 14, 15, 10, 22). Only one study of eight (12.5%) suggested that having a male child was associated with increased burden (2), and this study examined caregivers of children with leukemia. The authors suggested that this could be due to the fact that male gender is often related to increased incidence of behavioural difficulties (Boztepe et al., 2019).

Family size and siblings. Seven studies examined the link between family size or structure and burden. Five of the seven studies (71.4%) found no effect of these factors and carer burden when examining family size (1), being an only child (19), and the number of other siblings (2, 13, 21). One of the seven studies (14.3%) found that having a larger family was correlated with lower burden in caregivers (15), and one study (14.3%) suggested that having more children was associated with increased levels of burden (16). Additionally one study showed no link between burden and having a sibling with the same condition (8).

Ethnicity. Two studies examined ethnicity, and both studies (100%) found no difference based on the child’s ethnicity (7, 10).

Method of diagnosis. One study (100%) examined whether the method of diagnosis was associated with carer burden in caregivers of children with cystic fibrosis but found no evidence of this (8).

Illness severity. Fifteen studies examined the potential links between illness severity and burden, ten of the fifteen studies (66.7%) found that higher illness severity or disability factors predicted higher burden. Two studies found no relationship between burden and illness severity (6, 14) while four of these studies demonstrated a positive association between illness severity and carer burden (2, 9, 10, 12). A higher level of dependency and disability (15) and the child experiencing pain (7) were shown to be concurrent with increased burden across conditions. A number of studies examined disease specific severity factors. In caregivers of children with diabetes one study found that poor metabolic control was associated with increased burden (11), while another study found no evidence of this (13). This study also found no link found between carer burden and the number of tests, number of hospitalisations or the frequency of episodes (13). In caregivers of children with cancer, there was no link found with carer burden and the type of cancer, prognosis, or whether the child had relapsed or not (19). In caregivers of children with haemophilia there was no link found between carer burden and having a family history or a history of haem-arthrosis (21) or the type of haemophilia (6). However the number of bleeding episodes was linked to increased burden in caregivers of children with haemophilia (6). In caregivers of children with heart defects there was no difference in caregivers of children with Hypoplastic Left Heart Syndrome compared to Univentricular Heart Syndrome (17). The number or organs involved in cases of children with mitochondrial disease was correlated with increased burden for caregivers (18). In caregivers of children with cystic fibrosis testing positive for pseudomonas aeruginosa was related to experiencing higher burden while having meconium ileus appeared to have no effect on level of burden experienced by caregivers (8).

Illness duration and time since diagnosis. Six studies examined illness duration or time since diagnosis in relation to burden, and four of the six studies (66.7%) demonstrated no difference in carer burden based on time since diagnosis or duration of the child's illness or duration of caregiving (6, 13, 15, 19) while one study showed that more time since diagnosis was shown to be related to experiencing lower burden (5). This study examined caregivers of children with cancer, and it is possible that a longer time since diagnosis may be related to higher remission rates or a better prognosis. One study examining glaucoma showed that greater time since diagnosis was related to higher burden (23). These findings may be due to the fact that this is a degenerative disease where children may require increased support as time passes.

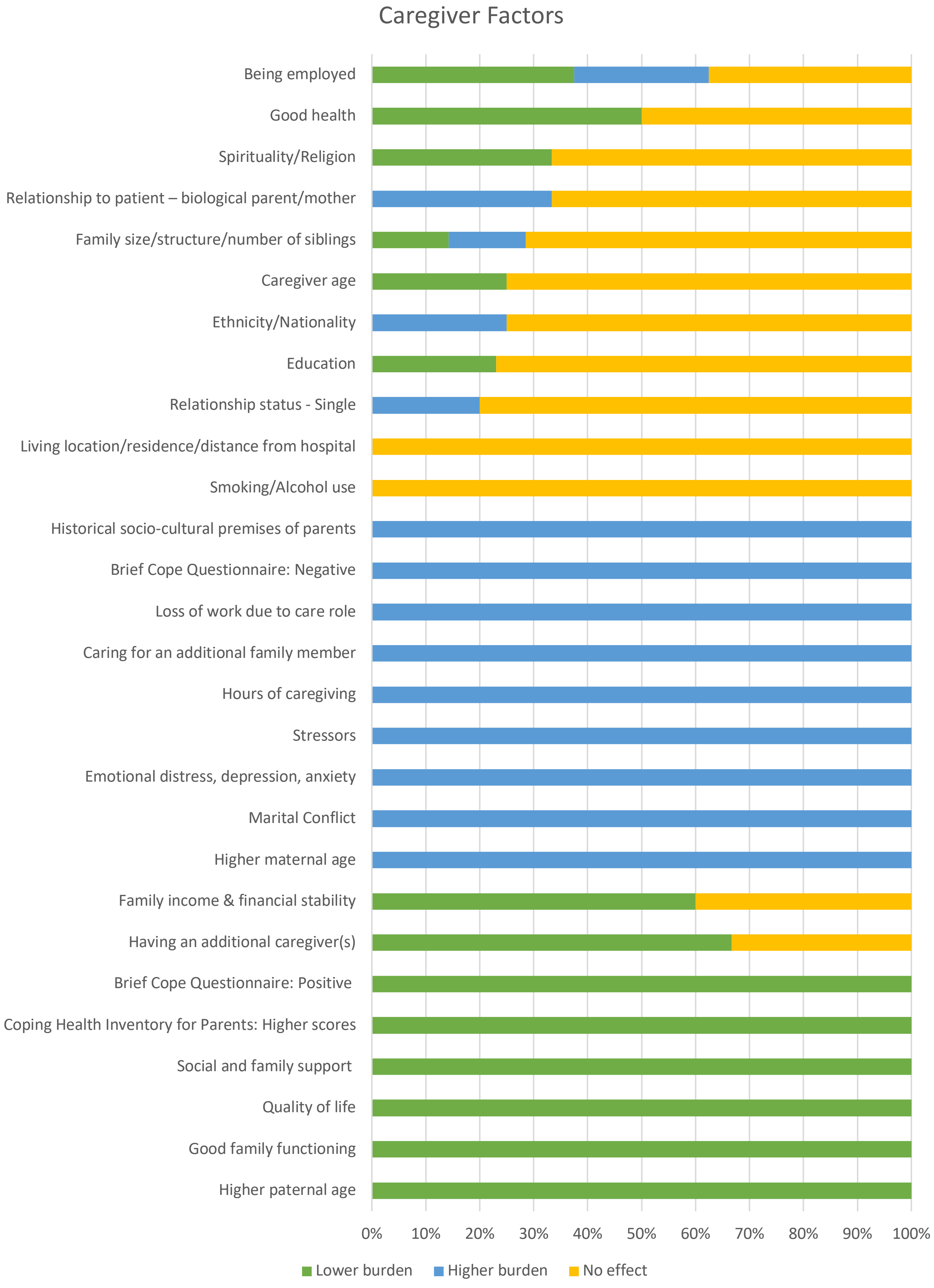
Treatment. Four studies examined treatment method or type and three of the four studies (75%) found no evidence of a link with burden. Three studies examined links with the number of treatments or appointments and two of the three studies (66.7%) found that having a higher number of hospital visits or treatments was associated with increased burden. One study examined cancer treatment severity and found no link (19). No relationship was found between carer burden and method of insulin administration in children with diabetes (13), using a port-o-cath or inhibitor (21) or gaining treatment via prophylaxis or not in children with haemophilia (6), or having cancer treatment in children with cancer (19). One study found using an inhibitor was linked with increased burden in children with haemophilia (6). In children with mitochondrial disease the number of hospital visits, number of speciality services included, and the number of medical visits were related to experiencing increased carer burden (18).

Comorbidities. Four studies examined whether having an additional illness was linked with caregivers experiencing higher burden and three of the four studies (75%) found evidence for this. Having an additional physical illness was associated with experiencing higher levels of burden in caregivers of children with heart disease (12) and Hypoplastic Left Heart Syndrome (3) but not cancer (2). The child experiencing internalising symptoms was associated with higher burden amongst caregivers of children with Irritable Bowel Disease (9). Additionally, two studies examined sleep and behavioural problems. Having sleep problems or behavioural difficulties were not related to caregiver burden (7), but behavioural and attention difficulties were related to increased burden in caregivers of children with heart disease (12).

Education. Three studies examined the impact of the child's education on carer burden and found no link (100%). Whether or not the child was in school, the child's level of education or their academic delay did not have an impact on caregiver burden (1, 14, 22).

Caregiver characteristics. A summary caregiver characteristics found to predict carer burden in caregivers is presented below. Figure 3 below represents a summary of these findings.

Figure 3. Graph displaying the percentage of studies showing evidence for links with lower, higher burden or no effect and caregiver factors.



Age. Eight studies examined caregiver age as a possible predictor of carer burden. six of the eight studies (75%) found no effect of child age on carer burden (6, 7, 13, 15, 19, 22). Two studies found that higher caregiver age acted as a protective factor (3, 18) and was correlated with lower burden. Additionally, higher caregiver age was associated with lower carer burden in carers of children with cancer (5) and higher maternal age was correlated with higher carer burden in caregivers of children with cancer (2).

Gender. Twelve studies examined gender. Nine of these twelve studies (75%) found no link between gender and burden (1, 3, 4, 6, 7, 14, 15, 20, 22). Only three studies indicated that being female was related to experiencing higher carer burden (8, 21, 23). However, most studies had a high percentage of female participants which may have resulted in difficulties with power (taking into account that most studies did not report their sample size calculations).

Ethnicity. Four studies examined caregiver ethnicity. Three of these four studies (75%) found no link between caregiver ethnicity and burden (3, 7, 22) while one study conducted in Brazil (16) suggested that being of a Black ethnicity was linked with higher burden.

Education. Thirteen studies examined education. Ten of these thirteen studies (76.9%) found no link between caregiver education level and burden (2, 3, 6, 7, 8, 13, 14, 15, 19, 22). Only one study suggested that higher education level could be a protective factor for carer burden (20) and two that lower education level would lead to higher burden (16, 23). This study (20) examined a mixed sample of childhood conditions and had a large sample size (although the authors failed to explicitly state sample size calculations).

Relationship status, conflict and family functioning. Five studies examined caregiver relationship status. Four of these five studies (80%) found no link between carer burden and relationship status (3, 6, 7, 22). One study suggested that being single was associated with increased burden in carers of children with cancer (19). Additionally, one study found a link between marital conflict and increased burden in caregivers of children with diabetes (11). Good family functioning was examined in two studies and both (100%) found that better family functioning was related to experiencing lower burden and poorer family functioning with higher burden (9, 20).

Relationship to the child. Three studies examined the relationship to the child. Two of the three studies (66.7%) found no difference in burden based on whether the caregiver is a mother or another caregiver (7, 11). Being the child's biological parent was linked to increased burden in caregivers of children with HIV (1) which could be explained by the incidence of HIV in parental caregivers in this sample.

Caregiver physical and mental health. Four studies examined whether caregiver health was associated with burden, and results were evenly distributed (50% suggested higher burden and 50% of studies showed no effect). Two of the four studies found no connection between carer burden and caregiver health (13, 15). One study found that having a chronic condition was not associated with experiencing more burden, but good health (measured using the SF36 subscales) was linked with lower burden (22). Another study found poorer caregiver health was correlated with increased burden (16). Additionally three studies examined distress and wellbeing in caregivers and showed that emotional distress (14), depression (22) and anxiety (20,22) were all associated with increased burden (100%). Wellbeing and quality of life were related to lower levels of burden while stressors were correlated with caregivers experiencing higher burden (20). One study also found no link between smoking or alcohol use (1).

Caregiving role. Three studies examined support with the caring role, and two of the three studies (66.7%) found that having help was related to lower burden. One study linked having an additional caregiver to assist with lower burden (22). One study examined additional support with caring and associated the lack of support with increased burden (16). One study found no impact of an additional caregiver on burden (2). Additionally, only one study examined hours of caregiving and found a connection between higher hours of caregiving and increased burden (22). Caring for an additional family member was also related to higher burden in one study (15).

Employment. Eight studies examined employment related factors and the results were split (37.5% showed links with lower burden, 25% higher burden, and 37.5% showed no effect). Three of the eight studies found no evidence of a link between carer burden and employment status (15, 19, 22). Two of the eight studies found positive relationships between carer burden and being unemployed (13, 16) and while one study found being employed was linked with decreased burden (7). This is as expected as being employed is largely correlated with financial stability which was also shown to influence burden. However one study showed links between being employed and lower levels of burden (23), and it is possible that this was related to increased pressures alongside the caring role. Additionally one study found that losing work due to a caring role was associated with increased burden (6) and a further study found a link between higher burden and with having a lower level of employment (17), both of which may link back to financial stability.

Family income and finances. Ten studies examined family income and finances and six of the ten studies (60%) showed that higher financial income was related to lower burden (6, 7, 15, 18, 22, 23). Four of the ten studies showed no link between finances and carer burden (2, 6, 10, 13).

Social support, coping and spirituality. Two studies examined social and family support and both (100%) found better support was related to lower burden (20,22). Two studies examined coping and both (100%) found that positive coping was linked with lower burden (4, 18). One study measured this with the Coping Health Inventory for Parents and found better family integration and support as well as coping was associated with lower burden (18). Additionally one study examined coping using the Brief COPE questionnaire (4). Coping was also connected to lower burden particularly when these coping strategies were positive such as positive reframing, acceptance, emotional support, and planning (4). When negative coping styles were used this was linked to higher burden including self-distraction, denial, substance use, behavioural disengagement, using emotional and instrumental support, self-blame and venting (4). The use of emotional support was associated with both lower and higher levels of burden across different dimensions of carer burden (4). Three studies examined spirituality and religion and two of these three studies (66.7%) found no link with burden and religion (1, 22) while one study showed a link with spirituality and lower burden (4). However these studies examined spirituality and religion in different ways such as using a structured tool (4) or asking demographic questions (1, 22).

Other. Five studies examined living location or distance from a hospital and all found no link with burden (100%). Living location (3, 13, 15, 22), or distance from a medical centre (6, 22) were not linked with burden. There also was no connection between carer burden and living in a multi-generational household (13). However experiencing family centred care was related to lower burden (5) and caregivers following historical socio-cultural premises was linked with higher burden specifically for caregivers in Mexico (20).

3.2.5 Unanimous Findings

From the studies examined in this review there were a large array of child/illness and caregiver characteristics identified. However, there were only a few factors in which all of the studies showed a unanimous directionality regarding the influence on caregiver burden which are changeable. Examining these factors specifically, and replicating findings across multiple studies and populations, might allow for the development of interventions to target these in order to reduce caregiver burden. It is worth noting that many of these factors were only examined by a small number of studies. These are displayed in Table 4 below.

Factors identified unanimously which are not changeable included higher paternal age being linked with lower burden (1 study), and higher maternal age, child premature birth and the historical-socio-cultural premise of caregivers being linked with higher burden (1 study each).

Factor	**Number of studies**
Factors linked with lower burden	
Good family functioning	2
Good caregiver quality of life	1
Having social and family support	1
Higher scores on Coping Health Inventory for Parents: Family integration/Social support/Total scores	1
Brief Cope Questionnaire: Positive reframing/Acceptance/Emotional support/Planning	1
Receiving family centred care	1
Factors linked with higher burden	
Emotional distress/depression/anxiety	3

Marital conflict	1
Experiencing stressors	1
Higher hours of caregiving	1
Caring for an additional family member	1
Loss of work due to caring role	1
Brief Cope Questionnaire: Self-distraction/Denial/ Substance use/behavioural disengagement/ Instrumental Support/Self-blame/Venting/Emotional support	1

4. Discussion

4.1 Summary of results

The current review summarises findings from studies examining factors which predict or are linked with carer burden in caregivers of children with chronic conditions. A total of 23 studies were included in this review. Assessment of quality was completed using the AHRQ risk of bias tool to critically evaluate methodological quality of included studies. The methodological quality of studies reviewed was mixed. Most importantly, the majority of studies failed to report whether they conducted power analyses, making it difficult to estimate whether or not studies had a high probability of encountering Type 2 errors.

The importance of this topic is well documented in previous research, highlighting that caregivers of children with chronic conditions experience elevated levels of mental health difficulties and distress (Pinquart, 2018) and lower quality of life and that these are related to higher levels of carer burden (Collins et al., 2020; Santo et al., 2011). In order to develop appropriate early intervention and support tools it is important to gain a better understanding of predictors of carer burden in this population. A previous review was completed in 2013 (Cousino et al., 2013) and examined pertinent literature up until 2012. The evidence base required an update, and the initial review also excluded the terms 'carer burden' or related concepts and focused instead on parenting stress. As such the review may have missed literature which referred to carer burden specifically. The current review synthesises findings from 23 studies published since 2012.

Previous models of caregiver burden have highlighted the importance of socioeconomic status, child functioning, caregiving demands, self-perceptions, social support, family functioning, and stress management in influencing caregiver burden in caregivers of children with cerebral palsy (Raina et al, 2004). Many of the factors felt to be influential in this model were also found to be linked with carer burden in the studies examined by this review. Unanimous results were found regarding the association of a number of factors with carer burden, although these findings came from a smaller number of studies. Of note were the findings that good family functioning and an absence of marital conflict was correlated with lower caregiver burden, and that the presence of emotional distress, anxiety and depression was associated with increased caregiver burden. Additionally, having a good quality of life, social support, using positive coping strategies and receiving family centred care were linked to experiencing lower caregiver burden. Contrastingly, using negative coping strategies, loosing work hours due to caring role, increased hours of care, experiencing stressors and caring for an additional family member were correlated with higher carer burden. These preliminary findings point towards the importance of supporting caregivers to improve their family functioning, coping strategies, quality of life and emotional wellbeing while also reducing the practical impact on carers (e.g. by reducing hours of care and providing support to reduce loss of work and financial difficulties). These factors are targetable with both social and psychological interventions, and so should be examined in future research.

Furthermore, the studies reported preliminary evidence suggesting that illness severity is associated with higher carer burden, contrary to findings from the review by Cousino and colleagues (2013) who suggested there was no evidence for such a link. Previous research also indicated a relationship between burden and illness severity of level of difficulties (Sales, 2003). In the present review over half of the studies examining this factor found evidence to suggest a connection or predictive value of the severity of the child's illness or disability. However a proportion of studies did not find a link. Both sets of studies were completed across different condition categories which were similar and used a mix of correlational and regression designs, and a mix of different measures so there was no apparent reason why findings would differ. However, all studies classified disease severity differently based on indicators of severity for each disease, which may account for the difference in results. For instance studies examining caregivers of children with diabetes based higher illness severity on worse metabolic control, number of tests, number of hospitalisations and frequency of episodes. In contrast, studies examining caregivers of children with cancer based severity on the type of cancer, the prognosis, and the incidence of relapse. Studies examining haemophilia categorised severity based on the type of haemophilia and the number of bleeds which studies of mitochondrial disease based this no the number of organs involved. These categories of severity also differed within the same disease across studies. For instance when looking at patients with cancer, Sulkers and colleagues (2015) examined severity as the type of diagnosis, prognosis, chance of survival and relapse while Boztepe and colleagues (2019) attempted to construct a scale to measure severity (which was unfortunately not accessible at the time of this review). Within studies, discrepancies were also found between different severity definitions, for instance Dekoven and colleagues (2014) found that having meconium ileus was linked with higher burden while the number of bleeding episodes showed no association with burden. As the studies used such a variety of definitions of disease severity despite overlap, comparing these studies is challenging without a generic measure for disease severity. Such a measure would have to also be illness specific in as far as possible while also assessing global parameters of illness severity. Future research may wish to examine whether measures exist or could be developed to examine illness severity across conditions, as was attempted by Boztepe and colleagues (2019) when comparing leukemia and cerebral palsy.

The present review also found that over half of included studies found that higher family income or financial stability was linked with lower burden in caregivers of children with chronic conditions. A proportion of studies found no effect of this however, and both groups of studies did not differ greatly in terms of considering a range of conditions or using a range of measures. The association with finances and income is not surprising as the construct of carer burden includes a subsection examining the financial consequences of the caregiving role. A number of measures of burden include questions relating to finances including the Zarit Burden Interview. Lower income and financial stability have been linked to higher carer burden and difficulties with the caring role in other informal caregiving populations such as those of elderly relatives (Williams et al., 2003; Yakubu & Schutte, 2018).

The majority of studies found no relationship between carer burden and caregiver education, caregiver gender, child gender, caregiver age, family size or structure, illness duration, caregiver relationship status or living location/distance from a hospital. However, these factors may be related to other risk factors identified, for instance the size or structure of the family might be related to family functioning or financial stability, or indeed social support or other caring responsibilities. Future research may wish to examine how these potential risk factors intercorrelate in order to build on theoretical knowledge and models of carer burden.

Two commonly explored factors showed mixed results in studies; child age and caregiver employment. The review showed that including half of the studies indicated no link between child age and burden, almost half indicated that higher child age relates to lower burden, and a small percentage showed higher age being associated with higher burden. Additionally, over a third showed no link between carer burden and employment, over a third showed lower burden when employed and a quarter showed higher burden with being employed. These studies examined a range of conditions and used a variety of measures of carer burden, suggesting that these findings could be generalisable across populations. However these results should be interpreted with caution however as there was no opportunity to compare effects, similarities and differences or confounding factors. Most studies also did not report power analyses so it is possible that no effects were found due to the fact that studies were underpowered. It may also highlight the importance of examining the individual caregiver's situation, for instance the type of employment may determine whether this is a protective or risk factor.

There are a number of issues which make the reviewing of findings around carer burden problematic. Firstly, carer burden is not always well conceptualised in literature (Bastawrous, 2013). Having such a variation in definitions of 'caregiver burden' may result in unclear findings which are hard to synthesise or evaluate (Bastawrous, 2013). There are studies which use the terms stress and burden interchangeably which may lead to confusion for readers. It is possible that the term 'burden' is seen as something very negative by caregivers, as they may feel it insinuates that their child is a burden on them. However this is a term used to describe a very specific type of stress experiences as a result of unique circumstances of a caring role and as such scientific studies and literature should try to be consistent in using the term burden so that findings can be more easily applied to practice and policy making.

Additionally, studies examining the concept of carer burden in this population continue to use a number of different measures of carer burden as well as integrating non burden measures of stress or distress which makes it difficult to compare findings and further confuses the concept of 'carer burden'. In the present review the majority of studies used the PIP and ZBI scales. However a range of other scales, specific subscales and abbreviated versions of measures were used. As such the summary of findings in this review should be interpreted with caution.

4.2 Implications for clinical practice

The current review presents a few preliminary suggestions of predictors of carer burden in caregivers of children with chronic conditions which may help inform screening and early intervention as well as support tool development. The most consistently found predictor of higher carer burden across studies included condition severity, having an additional illness and a higher number of hospital visits or procedures. These factors may determine the amount of support needed from the caregiver for daily tasks but also the risk to life for certain conditions e.g. degenerative or high risk conditions. These factors are also likely linked with higher hours of caregiving (for the child and others), loss of work due to caregiving role, and financial instability. While illness related factors may remain static, these factors could be targeted by providing additional practical support to families. These findings showed that having additional help with caregiving and that receiving family centred care were both linked with lower burden. Providing additional support for families may decrease burden, increase financial stability and promote the ability to remain at work.

A number of factors which might respond to targeted interventions were also identified in the review, albeit in a small number of studies. These included family functioning, quality of life, emotional wellbeing, social support, and coping strategies. When developing interventions to reduce carer burden, researchers and clinicians may wish to focus on enhancing caregivers ability to improve the above factors.

Other findings continued to be mixed even when a number of studies examined one aspect of child or caregiver characteristics. An overall recommendation which can be made at this time is that caregivers of children with chronic conditions should be monitored for carer burden during outpatient or hospital appointments, and support should be offered to them routinely. This support might initially take the form of information sharing about the potential effects of the caregiving role on carers which would allow for the normalisation of their experiences. Additionally, many carers turn to social media or peer support groups for information and help to cope with their feelings. It might be beneficial for clinicians to begin working jointly with these organisations or groups in order to develop appropriate peer led interventions for caregivers. Furthermore there is a need to involve caregivers more routinely as stakeholders in research and development in order to ensure interventions are co-produced and fir for purpose. This has been recommended by the Department of Health policy in the UK for some time (Boote et al., 2002), and seems to be lacking in research and intervention development in this population (Lopes-Vargas et al., 2019).

There are also many barriers to caregivers accessing avenues of support, which may be made more difficult by certain predictors indicated by this review such as financial instability and time constraints due to increased demands of the caring role when the child has a severe condition. A solution to this might be the introduction of blended care models which integrate e-health technologies such as online guided self-help into support packages for caregivers.

E-interventions are flexible and adaptable to the individual's experience and have been shown to be accepted by caregivers of children with chronic conditions (Palermo et al., 2009). The use of technology may be particularly useful for caregivers who have little support with the caring role, have other caring duties or are unable to access other forms of traditional support for a variety of reasons.

4.3 Future research

The review by Cousino and colleagues (2013) also evidence for a role of found greater parental responsibility for treatment management and lower levels of child self-care. These factors were not examined by studies included in this review, but future research may wish to replicate these findings. Future studies examining predictors of carer burden should be mindful of the quality and risk of bias of their research, and ensure they control for confounding variables as well as report their power analyses in order to be transparent about their findings.

It is difficult to draw conclusive evidence from research examining predictors of burden without examining effect sizes or combinations of potential predictors (it is likely that a number of predictors will conflate to increase burden rather than affecting burden individually). Future research may wish to examine these predictors in a more rigorous manner, by examining effect sizes or associations between various predictors. This was not within the scope of the present review. Additionally, the role of caregiving for a child with a chronic condition is a complicated and ever changing experience. While reviews like this can attempt to synthesis the information presented by quantitative studies, despite the challenges of this described above, it is important to acknowledge that each caregiver will be unique. Future research may wish to integrate the viewpoint of caregivers when examining predictors of carer burden. Using qualitative or mixed designs will allow the researcher to gain a broader view of predictors of carer burden, as suggested by Bastawrous (2013).

4.4 Quality and Limitations of the Current Review

The current review followed the recommendations from the AMSTAR quality checklist which is suitable for systematic reviews (Shea et al., 2007). As such, an *a priori* design was registered on Prospero, a comprehensive literature search was conducted, all studies from grey literature sources were examined to ensure they were peer reviewed, a proportion of data was extracted by a second independent rater to reduce bias, and the scientific quality was assessed using a structured tool by the main author (58% of which was blindly double assessed by two independent raters).

However, there are a number of limitations of the current review, and the results presented herein should be interpreted with these in mind. The heterogeneity of outcome measures used meant that synthesis of findings and comparison of results was challenging. Similarly, the disparity in ways of conceptualising various predictors such as illness severity added to this complication. While this review purposefully included all physical chronic conditions, the wide spread of condition types also added to the challenge of directly comparing results.

While the review searched a wide range of databases including grey literature and examined reference lists of all included studies, it is possible that the range of databases searched lead to the loss of potentially eligible studies. This review also excluded studies examining neurodevelopmental or mental health conditions or mixed samples. Future research may wish to include these samples in reviews in order to be able to comment on a wider range of childhood conditions. It would also be interesting to review whether there are differences in findings based on the type of condition (i.e. physical chronic conditions compared with neurodevelopmental or mental health conditions).

4.5 Conclusion

The current review summarized predictors of carer burden in caregivers of children with chronic conditions. The results highlight preliminary findings about potential predictor variables, which need to be further examined in a larger sample using standardised measures of carer burden and ensuring analyses are adequately powered. The evidence base clearly highlights increased burden in caregivers of children with chronic conditions, and there is a need to continue to explore factors which may be linked to this so that appropriate early interventions and support tools can be developed to target those caregivers who may be particularly vulnerable to experiencing carer burden and its consequences.

5. Included Studies

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Appendix A. Data Extraction Form

Table A1

Sample Data Extraction Form

Study (Year, Location, Authors, Child Conditions)	**Study design**	**Sample characteristics**	**Inclusion/ Exclusion criteria**	**Carer burden/ parental stress measures**	**Other measures**	**Relevant findings**	**Predictors of carer burden**

Appendix B. Measures used in the included studies

Table B1

Measures used in the included studies

Instrument	Number of studies	Format	Subscales
Paediatric Inventory for Parents	7	5 point Likert scale with 42 items	Difficulty and frequency: Communication, emotional functioning, role functioning and medical care.
Zarit Burden Inventory	6	5 point Likert scale with 22 items	None
Caregiver Burden Inventory	2	4 point Likert scale with 24 items	Physical, social, emotional, time-dependence and developmental burden
CarerQoL	2	3 point Likert scale with 7 items and a VAS scale examining happiness	None
Caregiver Burden Scale	2	Likert scale with 22 items	General strain, social isolation, environment, emotional involvement, and disappointment
Parenting Stress Index – Short Form	2	4 point Likert scale with 36 items	Parental Distress, Parent-Child Dysfunctional Interaction, and Difficult Child
Impact on Family Scale (15 items)	1	4 point Likert scale with 15 items	Subset of a larger 24 item questionnaire
Caregiver Strain Index	1	4 point Likert scale with 13 items	None
Objective Burden subscale of the Revised Burden Scale	1	5 point Likert scale with 6 items	This is a subscale of a larger questionnaire
Parenting Stress Index	1	4 point Likert scale with 101-item	Child domain and parent domain
Positive Aspects of Caregiving	1	Likert scale with 9 items	Self-affirmation and outlook on life

Chapter 2. Empirical Study

The relationship between carer burden, self-compassion, psychological flexibility and wellbeing in caregivers of children with chronic conditions

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Abstract

Aim: This study examined the relationships between carer burden, self-compassion, psychological flexibility and wellbeing in caregivers of children with chronic conditions. Higher levels of carer burden and mental health difficulties have been evidenced in this population, and the current study examined whether carer burden, self-compassion and psychological flexibility predicted wellbeing. **Design:** Participants (*N* = 205) were recruited via social media, and completed an online survey encompassing demographic information and measures of burden (ZBI), self-compassion (SCS), psychological inflexibility (AAQ-II), anxiety (GAD-7), depression (PHQ-9) and quality of life (QoLS). **Results:** In combination carer burden, self-compassion and psychological inflexibility all significantly predicted anxiety, depression and quality of life with large effect sizes. Carer burden was found to be a predictor of higher anxiety and lower quality of life but did not predict depression. Psychological inflexibility predicted higher anxiety and depression scores and lower quality of life. Self-compassion predicted lower anxiety and depression scores and higher quality of life. **Conclusion:** Carer burden, psychological inflexibility and self-compassion appear to be important targets for intervention to improve wellbeing in caregivers of children with chronic conditions.

Keywords: Carer Burden, Self-compassion, Psychological Inflexibility, Anxiety, Depression, Quality of Life

Abbreviations
ZBI – Zarit Burden Interview
SCS – Self-compassion Scale
AAQII – Acceptance and Action Questionnaire II
GAD7 – General Anxiety Disorders Assessment 7
PHQ9 – Patient Health Questionnaire 9
QoLS – Quality of Life Scale

1. Introduction

Childhood chronic conditions are defined as illnesses lasting or expected to last at least three or recurring (Mokkink et al., 2008). The prevalence of long-term conditions in childhood has been increasing (Liu et al., 2015); between 15 % and 27% of children and adolescents are estimated to be impacted (Van der Lee et al., 2007; Van Cleve et al., 2010), and it is predicted that this number will continue to rise with further advances in medical interventions and increased survival rates (Liu et al., 2015). The psychological impact of childhood disease on the child has been well documented, including higher rates of psychological comorbidities (Coburn et al., 2019) and emotional and behavioural problems (Pinquart & Shen, 2011). However the impact of childhood chronic illness is widespread including an effect on the wider family system (Cousino & Hazen, 2013). For instance, research has demonstrated reduced family functioning in families of children with perinatal stroke (Bemister et al., 2014) and Autism Spectrum Disorder (Karst & Van Kecke, 2012), as well as family disruptions and role changes in families of children with diabetes (Whittemore et al., 2012) and cancer (Long & Marsland, 2011; Jones, 2012).

1.1 Caregiver mental health

As well as impacting the wider family system, the experience of raising a child with a chronic condition has been shown to adversely impact primary caregiver's wellbeing. Above population norm levels of distress and mental health difficulties have been identified in caregivers of children with chronic illnesses (Pinquart, 2018) including caregivers of children with asthma (Kopel et al., 2017), cystic fibrosis (Brucefors et al., 2015), diabetes (Hessler et al., 2016) and epilepsy (Reilly et al., 2018) among others. Caregivers of children with a chronic illness have been found to have higher levels of depression and anxiety (Besier et al., 2011; Van Oers et al., 2014), higher rates of burnout (Lindstrom et al., 2010), higher levels of perceived stress (Masa'Deh et al., 2012; Marsland et al., 2013), lower levels of self-confidence, self-efficacy, sociability and wellbeing (Barlow & Ellard, 2006) and lower levels of quality of life (Lawoko & Soares, 2003).

Gaining a better understanding of caregiver distress and mental health in this population is crucial as previous findings have shown that caregiver's wellbeing can impact on child functioning. Whittemore and colleagues (2012) found that caregiver emotional distress was related to higher child stress, lower child quality of life, and difficulties with diabetes management. Caregivers whose children have a chronic condition seem to experience higher stress than caregivers of children without a chronic condition (Cousino et al., 2013). The stress specific to caregiving has been termed carer burden or caregiving stress, and in this population is often referred to as parental burden or stress.

1.2 Carer Burden

Carer burden has been defined as the level of stress (Pian et al., 2017), as well as the physical, psychological, social, relational, and financial problems experienced by caregivers as a result of their caring role (Whalen & Buchholz, 2009). Carer burden is a distinct concept from global or generic stress as it focuses on the impact of the caring role on the caregiver while stress relates to the feeling of tension experienced by an individual when the environment is perceived as taxing or dangerous (Butler, 1993). Carer burden has been associated with a number of mental health difficulties in carers. In caregivers of children with cancer, higher carer burden has been found to be negatively correlated with mental health (Santo et al., 2011). Carer burden has also been linked to increased anxiety and depression in caregivers of children with intellectual disabilities (Gallagher et al., 2008) and lower quality of life in caregivers of children with life limiting conditions (Collins et al., 2020). Further research is warranted to examine whether carer burden could predict wellbeing in this population.

Studies from caregivers of adult patients also suggest an association between higher carer burden and higher perceived stress, for instance in caregivers of adults with cancer (Ramos-Campos et al., 2020), elderly spousal caregivers (Luchesi et al., 2016; Gratao et al., 2019), and carers of adults with schizophrenia (George & Raju, 2006). Watson (1997) found higher perceived stress scores amongst caregivers of children with renal replacement therapy who also had higher are burden levels, suggesting a potential relationship between level of carer burden and perceived stress in this population. However, there has been little recent research on the link between carer burden and perceived stress in caregivers of children with a range of chronic conditions. As such further research is needed to update the evidence base, to enhance the understand this association in caregivers of children with chronic conditions, and to aid in the development of prevention and interventions tools.

1.3 Interventions addressing carer burden in caregivers of children with chronic conditions

From the limited existing research, preliminary results suggest that psychological interventions which directly target caregiver mental health (Eccleston et al., 2012) and problem-solving approaches are effective in alleviating distress (Wade et al., 2006; Palermo et al., 2016; Eccleston et al., 2015). However, the majority of interventions for caregivers of children with chronic conditions appear to focus on outcomes including child symptoms and caregiver disease related behaviours e.g. checking the child's health measurements. Only a small number of studies measured carer burden specifically as an outcome in interventions for caregivers of children with chronic conditions, and the tools used to measure this are varied.

A number of studies assessed parenting stress using the Parenting Stress Index (PSI). Tsiouli and colleagues (2014) found benefits of progressive relaxation and breathing techniques over 8 weeks in caregivers of children with Type 1 diabetes in decreasing parenting stress or perceived stress. Neece (2014) found reductions in parenting stress in caregivers of children with developmental delays following a mindfulness-based stress reduction intervention. Palermo and colleagues (2016) found differences in caregiver stress at follow up with a problem-solving intervention for caregivers of children with chronic pain. However, there seems to be little evidence whether these effects are maintained over time. Despite initial evidence suggesting that problem solving interventions improve caregiver wellbeing (Eccleston et al., 2015), Delve and colleagues (2006) found no decrease in parenting stress at follow up with a problem-solving competence-based intervention for caregivers of children with rare conditions. It is possible that caregivers of children with rare conditions may experience less support, and that comorbid behavioural difficulties in these children increases the stress experienced by caregivers.

A number of studies used generic measures such as the Perceived Stress Scale (PSS) or defined parenting stress as a combination of anxiety or depressive symptoms e.g. using the Depression-Anxiety-Stress Scale (DASS) rather than focusing on formal measures of caregiver burden. These studies appeared to find no effect of interventions on stress in caregivers, which suggests the need for a distinction between generic stress and parenting stress and the use of formal measures of burden. Marsland and colleagues (2013) found no significant decrease in stress levels measured using the PSS following a stress management intervention at 4-5 months post diagnosis in caregivers of children with cancer. Stehl and colleagues (2009) similarly found no effect of a family competency intervention at follow up on caregiver traumatic stress using the Acute Stress Disorder Scale (ASDS). Sassman and colleagues (2012) found no difference in the DASS stress component at 6 and 12 months following parenting skill and education intervention.

The results of the studies above suggest that specific measures of carer burden should be used such as the Zarit Burden Interview (ZBI) or the Parenting Stress Index (PSI). Whalen and colleagues (2009) report that the ZBI is most commonly used in studies examining carer burden, and a number of studies have adapted this tool and used it in populations of caregivers of children with chronic conditions (Javalkar et al., 2017; Asadullah et al., 2017; Wang et al., 2017; Boztepe et al., 2019; Pruthi & Mohta, 2010; Pruthi & Singh, 2010). Using standardized measures when researching such concepts allows for results to be compared across studies and ensures that we refer to a specific construct rather than loosely identifying stress across studies by measuring potentially different outcomes.

Research priorities identified by clinicians and caregivers of children with chronic conditions included developing support for caregivers and reducing carer burden (Lopez-Vargas et al., 2019). Further research is needed to develop and evaluate interventions targeted specifically at carer burden in caregivers of children with chronic conditions. As such it seems imperative to better examine the mechanisms of change for reducing carer burden in caregivers of children with chronic conditions. Two factors which have been identified from previous studies are self-compassion and psychological flexibility.

1.4 Self-compassion

Self-compassion is defined as being kind to oneself and consists of three main components: a) self-kindness rather than self-judgement, b) viewing experiences as a part of the human experience rather than isolating and c) mindfully holding painful thoughts rather than overidentifying with them (Neff, 2003). Research has demonstrated that higher levels of self-compassion are associated with lower levels of depression, anxiety and stress (Marsh, Chan,

& MacBeth, 2018; Krieger et al., 2013; Pinto-Guoveia et al., 2014; Barnard & Curry, 2011) and higher wellbeing (Keng & Liew, 2016). A systematic review has found strong negative associations between self-compassion and psychopathology (anxiety, depression and stress) across age groups (MacBeth & Gumley, 2012), and it appears that self-compassion may act as a protective factor against distress (Barnard et al., 2011; Keng & Liew, 2016). This suggests a potential role for self-compassion in caregivers of children with chronic conditions, who have been shown to experience high levels of distress (Pinquart, 2018).

Self-compassion has indeed been associated with reduced caregiver distress and psychopathology in caregivers of children with chronic conditions. In caregivers of children with autism higher levels of self-compassion were linked with lower levels of caregiver distress and depression regardless of the severity of child symptoms (Neff & Faso, 2015). Self-compassion has been thought to moderate the effect of stress on mental health by activating a sense of safety within the individual and allowing for better emotion regulation (Hickey et al., 2017). Alongside the influence of self-compassion on mental health, research indicates that there is a relationship between self-compassion and carer burden which suggests that higher levels of self-compassion act as a protective factor. In caregivers of individuals with dementia, higher levels of self-compassion have been associated with lower levels of carer burden (Lloyd et al., 2019).

There are a few studies which have examined the relationship between self-compassion and carer burden in caregivers of children with chronic conditions. In caregivers of children with autism, self-compassion was negatively related to caregiver stress (Neff & Faso, 2015), and the higher scores in the negative dimensions on the self-compassion scale (SCS) were related to higher caregiver stress (Bohadana et al., 2019). Self-compassion was also found to be associated with lower levels of stress and depression in caregivers of children with intellectual and developmental disabilities (Robinson et al., 2018), and correlated positively with wellbeing and quality of life in caregivers of children with diabetes (Hammer, 2014). These studies evidenced a connection between higher self-compassion and lower carer burden using formal measures of burden (PSI), which ensures that they are measuring the discrete concept of ‘carer burden’ or ‘parenting stress’ rather than a generic measure of stress or wellbeing. However these studies seem to be mostly concentrated in the population of caregivers of children with neurodevelopmental conditions. As such further up to date research is required to examine whether this apparent protective nature of self-compassion extends to caregivers of children with other chronic conditions, and how self-compassion may be involved in the link between carer burden and wellbeing in caregivers.

There is some evidence that self-compassion may moderate the relationship between carer burden and wellbeing. Hickey and colleagues (2017) found that self-compassion moderates the impact of stress on depression. Hsieh and colleagues (2019) found that higher self-compassion was found to be associated with less depressive symptoms in caregivers of adult patients with lung cancer. Additionally, self-compassion was found to moderate the effect of caregiving stress on depressive symptoms. As there is little evidence of this in the population of caregivers of children with chronic conditions, it would be beneficial to examine whether self-compassion predicts caregiver wellbeing in this population and across conditions.

Gaining a better understanding of the potential mechanisms of change will allow for the development of prevention and intervention techniques to support caregivers of children with chronic conditions and reduce the impact of carer burden. While self-compassion based interventions do not seem to have been widely used in caregivers of children with chronic conditions, it has been demonstrated to be a mechanism of change in other populations. Interventions targeting self-compassion have been shown to reduce depressive symptoms (Barnard & Curry, 2011). A review by Wilson and colleagues (2019) showed that self-compassion related therapies such as compassion-focused therapy (CFT), mindfulness based cognitive therapy (MBCT) and acceptance commitment therapy (ACT) were effective in increasing self-compassion and reducing anxiety and depression. This suggests that self-compassion may act a potential agent of change for individuals experiencing anxiety and depression. Therefore, it is reasonable to suggest further investigation into the role of self-compassion in this population as a precursor to intervention development.

1.5 Psychological flexibility

Another potential mechanism of change in reducing psychological distress is psychological flexibility, which is defined as the ability to experience the present moment consciously and change behaviour when appropriate (Hayes et al., 2006). Lower levels of psychological flexibility have been showed to coincide with increased psychopathology including depression and anxiety in the general population (Kashdan & Rotternberg, 2010). Preliminary research indicated an important role for psychological flexibility in caregivers of children with chronic conditions. Specifically, Sairanen and colleagues (2018) showed that lower levels of psychological flexibility predicted caregiver distress (anxiety, depression, stress and burnout) in caregivers of children with chronic conditions. Slowey (2014) examined informal carers and found that higher levels of psychological inflexibility were associated with higher levels of distress and lower wellbeing, and psychological inflexibility was found to moderate the relationship between distress and wellbeing but not between burden and wellbeing. While there is initial evidence for the buffering effect of psychological flexibility in the development of caregiver distress, there is a need to examine whether psychological inflexibility could act as a robust predictor of wellbeing in this population.

An intervention which aims to increase psychological flexibility is Acceptance and Commitment Therapy (ACT) (Hayes et al, 2006). For caregivers of children with autism ACT based interventions have been shown to reduce depressive symptoms (Blackledge & Hayes, 2006), reduced anxiety and depression as well as improved quality of life (Poddar et al., 2015). In caregivers of children with life-threatening illness ACT based interventions have been found to be effective in reducing post-traumatic stress symptoms and the emotional impact of the child's illness (Burke et al., 2014). Psychological flexibility has been shown to correlate with self-compassion and both factors predicted emotional wellbeing in a non-clinical population (Marshall & Brockman, 2016). There is limited research examining psychological flexibility in caregivers of children with chronic conditions. Previous studies suggest that it this may influence caregiver mental health and wellbeing and act as a mechanism for change in interventions. However further research is needed to examine whether this also applies for caregivers of children with chronic conditions and whether psychological inflexibility may predict wellbeing in this population.

1.6 Social Media Recruitment

As detailed above, this study population are likely to experience increased demands, mental health difficulties and carer burden due to their caring role. Caregivers often use online platforms to search for support for their child and themselves, and as such social media platforms provide a good forum for recruitment. Social media has become increasingly popular for study recruitment as it allows researchers to read a wide pool of potential participants who can be identified to meet inclusion criteria based on shared personal information such as group membership (Gelinas, Pierce, Winkler, Cohen, Lynch & Bierer, 2017). There is evidence for the efficacy of social media recruitment for hard to reach populations such as patients with rare diseases, stigmatised or minority groups and those with mental health difficulties (Gelinas et al., 2017). Additionally, social media recruitment allowed the researchers to contact a number of international charities and organisations supporting children or caregivers, in order for these to promote the study through their social media presence.

However, it is likely that utilising social media platforms for recruitment will result in selection bias for those who choose to interact with the survey, and will exclude caregivers who do not have access to technology for instance those without internet access due to sociodemographic or living environment factors. Additionally, there are risk in terms of participant communication on social media regarding both direct communication with the researcher and with other participants (Gelinas et al., 2017). However this also allows researchers to correct misperceptions and answer questions about the study easily, and make answers publicly available thereby reducing demand on both the research team and participants.

1.7 Study aims and hypotheses

This study examined the associations between carer burden, psychological inflexibility and self-compassion and caregiver wellbeing (anxiety, depression, stress and quality of life) in caregivers of children with chronic conditions. The study also examined whether carer burden, self-compassion and psychological flexibility might predict caregiver wellbeing.

The study hypotheses are as follows:

1) Higher carer burden will be associated with higher psychological inflexibility, anxiety, depression, and stress, and lower quality of life and self-compassion in caregivers.
2) Higher self-compassion will be associated with lower carer burden, anxiety, depression, and stress, and higher quality of life in caregivers.
3) Higher psychological inflexibility scores will be associated with higher carer burden, anxiety, depression, and stress, and lower quality of life in caregivers.
4) Carer burden, self-compassion, stress and psychological inflexibility will predict caregiver wellbeing (anxiety, depression, stress and quality of life). It is predicted that

self-compassion will have a positive relationship with quality of life and a negative relationship with anxiety, stress and depression. It is hypothesized that carer burden and psychological inflexibility will have a negative relationship with quality of life and positive relationships with anxiety, depression and stress.

2. Methodology

2.1 Study Design

This study used a cross sectional design to quantitatively examine the relationship between carer burden, self-compassion, psychological inflexibility and wellbeing in caregivers of children with chronic conditions.

2.2 Ethical Approval

Ethical approval was granted by the School of Health in Social Science Research Ethics Committee and sponsorship was obtained from the University of Edinburgh on October 14th, 2019. Please see Appendix A.

2.3 Participants

This study recruited caregivers of children with chronic health conditions. The term caregiver herein refers to all participants (including parents and other caregivers such as family or foster carers). A chronic condition was defined as any condition (physical, mental or neurological) which lasts or is expected to last 12 months or longer. To be eligible for the study, participants were required to fulfil the following inclusion criteria: i) aged 16 or over, ii) self-identified to be fluent in English, iii) provided informed consent following the reading of an information leaflet. Participant data was excluded if they did not appear to be able to answer the questions in English or were aged 16 or under, or if their child was over the age of 18 years.

A total of 319 individuals clicked on the survey webpage, and 219 consented to taking part in the study and completed the survey fully indicating a response rate of 68.7%. A number of participants were excluded from the analyses ($N = 14$) due to the fact that their child was over the age of 18, and the final sample therefore consisted of 205 participants; demographic data of the sample are presented in Table 1 below. The sample of caregivers was mostly female (95%) which is common in participant samples in this population as demonstrated in the systematic review (Schroeter, 2020) which showed an average of 76% female participants in studies examining carer burden in parents of children with chronic conditions. This limits the generalisability of findings to male caregivers.

Table 2 displays the demographic data of their children with chronic conditions. The prevalence of specific chronic conditions in the sample of children whose caregivers took part in the study can be found in Appendix F (Table 11). The most common conditions in the children of the caregivers were kidney or bladder disorders (20%), digestive system disorders (19%), anxiety (14.1%) and autism spectrum disorders (13.7%). Over half of the sample had a child with more than one health condition (52.2%). Of those conditions reported by caregivers the most common were physical (64%), followed by neurodevelopmental (18.3%) and mental or emotional disorders (17.6%).

Table 1

Demographic Characteristics of Participants

Demographic variables	*N* (%)
Age range	
16 – 25 years old	1 (0.5%)
26 – 50 years old	180 (87.8%)
51 – 65 years old	23 (11.2%)
Missing	*1 (0.5%)*
Gender	
Female	195 (95.1%)
Male	5 (2.4%)
Missing	*4 (2%)*
Ethnicity	
White	95 (46.3%)
Mixed	76 (37.1%)
Asian	8 (3.9%)

Black	12 (5.9%)
Latin	6 (2.9%)
Other	6 (2.9%)
Missing	*2 (0.98%)*
Relationship Status	
Single	13 (6.3%)
In a relationship	24 (11.7%)
Married	138 (67.3%)
Widowed	2 (1%)
Separated/Divorced	27 (13.2%)
Missing	*1 (0.5%)*
Employment status	
Unemployed	25 (12.2%)
Part-time	56 (27.3%)
Full-time	76 (37.1%)
Other (including carer, disabled, student, self-employed etc.)	46 (22.4%)
Missing	*2 (1%)*
Education level	
University or college	134 (65.4%)
Between secondary and university	42 (20.5%)
Secondary school	27 (13.2%)
Primary school or less	1 (0.5%)
Missing	*1 (0.5%)*
Country of residence	
United Kingdom	95 (46.3%)
United States of America	76 (37.1%)
Australia	8 (3.9%)
Canada	12 (5.9%)
Europe	6 (2.9%)
Asia	3 (1.5%)
South America	3 (1.5%)
Missing	*2 (1%)*
Living location	

In a town or city	92 (44.9%)
In the suburbs	70 (34.1%)
In the countryside	42 (20.5%)
Missing	*1 (0.5%)*
Number of other children (Range 0 – 7)	
None	27 (13.2%)
1	80 (39%)
2	58 (28.3%)
3	24 (11.7%)
4 or more	15 (7.3%)
Missing	*1 (0.5%)*
Number of other children who have a chronic condition	
None	115 (56.1%)
1	48 (23.4%)
2	14 (6.8%)
3	3 (1.5%)
4	2 (1%)
Missing	*23 (11.2%)*
Previous mental health difficulties before child’s diagnosis	
No	122 (59.5%)
Yes	82 (40%)
Missing	1 (0.5%)
Own health conditions	
No	100 (48.8%)
Yes	103 (50.2%)
Missing	*2 (1%)*

Table 2

Demographic Characteristics of Children with Chronic Conditions

Demographic variables	*N* (%)
Age (Range 0 -18)	
0 – 5 years old	33 (16.1%)

6 – 10 years old	50 (24.4%)
11 – 15 years old	83 (40.5%)
16 – 18 years old	39 (19%)
Type of condition(s)	
Physical	106 (51.7%)
Mental	17 (8.3%)
Neurodevelopmental	42 (20.5%)
Physical and mental	8 (3.9%)
Physical and neurodevelopmental	12 (5.9%)
Mental and neurodevelopmental	11 (5.4%)
Physical, mental and neurodevelopmental	6 (2.9%)
Other	2 (1%)
Missing	*1 (0.5%)*
Number of conditions (Range 1 – 9)	
1	97 (47.3%)
2	50 (24.4%)
3	27 (13.2%)
4 or more	30 (14.63%)
Time since diagnosis (Range 0 – 15)	
Less than 1 year	14 (6.8%)
1 – 3 years	76 (37.1%)
3 – 10 years	87 (42.4%)
10 – 15 years	26 (12.7%)
Missing	2 (1%)

2.4 Procedure

The questionnaire was pilot tested by four adult volunteers, one of whom had a chronic health condition and three of whom were caregivers of children with chronic conditions. This allowed for feedback on questionnaire design and for an estimated completion time to be calculated. Participants were recruited between September 2019 and January 2019 via advertisements on social media including via a dedicated Twitter account and on Facebook groups for caregivers of children with chronic conditions. When recruiting from social media groups, the researcher contacted administrators of Facebook groups privately with a description of the study and a link to view the questionnaire, and requested permission to post this onto the page. Once approval was provided, the researcher posted the description and link onto the page. When tweeting organisations or asking for the survey to be shared, the researcher messaged relevant administrators to ask for their support or tagged them in a post. The researcher interacted with comments from individuals relating to the post, for instance to clarify whether certain conditions would be included or what the duration of the survey was. The study was constructed using Qualtrics, an online survey building tool. Once participants logged onto the website they were asked to read an information leaflet and complete a consent form (Appendix B). Following the completion of the survey participants were presented with a debrief form (Appendix C).

2.5 Measures

Caregivers and carers of children with chronic conditions completed an online questionnaire which included demographic questions (Appendix D) as well as a battery of questionnaires containing measures relating to mental wellbeing, self-compassion, and psychological inflexibility (Appendix E). The measures were presented in the order below.

2.5.1 Demographics

Caregivers were asked to complete a number of questions regarding their and their child's demographic information including age, gender, relationship status, disease factors, socioeconomic factors, and family factors (see Appendix D).

2.5.2 Zarit Burden Interview

While there are a number of carer burden measures used in literature, a review by Whalen and colleagues (2009) suggested that the 22-item Zarit Burden Interview is the most widely used. The 22-item questionnaire was originally developed to assess carer burden in caregivers of patients with dementia (Zarit et al., 1980) and was modified for this study by replacing the term relative to child(ren) in each question. Similar modifications have been used previously in studies examining carer burden in caregivers of children with chronic illnesses (Javalkar et al., 2017). While further psychometric testing of carer burden measures needs to be conducted for populations such as caregivers of children with chronic conditions (Whalen et al., 2009), the Zarit Burden Interview has been used in a number of studies examining carer burden in caregivers of children with chronic conditions including HIV (Asadullah et al., 2017), leukemia (Wang et al., 2017; Boztepe et al., 2019), anorectal malformations (Pruthi & Mohta, 2010), cerebral palsy and thalassemia (Pruthi & Singh, 2010).

Scores are totalled to calculate a total score, and higher scores indicate higher carer burden for caregivers. Scores can be categorised according to the level of severity, including little or no burden (0 – 21), mild to moderate burden (21 – 40), moderate to severe burden (41 – 60) and severe burden (61 – 88). The ZBI has been shown to have good internal consistency reliability (Cronbach's alpha coefficient of .92) and was not related to age, gender, relationship status or employment in the sample examined which suggests that it is a good measure across different populations (Herbert, Bravo & Preville, 2000). In the current study, internal consistency was good ($\alpha = 0.89$).

2.5.3 Acceptance and Action Questionnaire II

The Acceptance and Action Questionnaire II (AAQ II) measures psychological inflexibility. The 7 questions with Likert scales are scores are summed to gain a total score. The questionnaire has been shown to have good reliability and validity (alpha coefficient of 0.84) and good test-retest reliability with 0.81 and 0.79 respectively (Bond et al., 2011). The scale showed excellent internal consistency in the present study ($\alpha = 0.92$).

2.5.4 Self-compassion Scale

The 26 item Self Compassion Scale (SCS) measures self-compassion. Likert scale scores are summed to provide a total score after reverse scoring where applicable. While the scale allows for subscales to be calculated; in this study, only the total score was used as there were no hypotheses specific to individual subscales. The scale has been shown to have good internal consistency (Cronbach's alpha of 0.92) and is theoretically valid and psychometrically reliable (Neff, 2003). Internal consistency for this scale was excellent in the present study ($\alpha = 0.90$).

2.5.5 Generalized Anxiety Disorder Assessment 7

The Generalized Anxiety Disorder Assessment 7 (GAD7) measures symptoms of anxiety. The scores are summed to form a total score which is classified according to cut-off scores indicating severity levels of systems, from mild (0 – 5), moderate (6 – 10), moderately severe (11 – 15) to severe anxiety symptoms (16-21). The 7 item scale has been shown to have good internal consistency (Cronbach's alpha of 0.89), reliability and validity in measuring anxiety in the general population (Lowe et al., 2008). The scale demonstrated excellent internal consistency in this study ($\alpha = 0.91$).

2.5.6 Brief Patient Health Questionnaire 9

The Brief Patient Health Questionnaire (PHQ9) measures symptoms of depression. The scores are totalled to compute a total score, which are classified into severity scores of mild (0 – 5), moderate (6 – 10), moderately severe (11 – 15) and severe depressive symptoms (16 – 20). The 9 item scale has been shown to have good internal reliability (Cronbach's alpha of 0.89), good test-retest reliability (0.89) and satisfactory criterion, construct and external validity (Kroenke et al., 2001). This scale had good internal consistency in this study ($\alpha = 0.87$).

2.5.7 Perceived Stress Scale

The Perceived Stress Scale (PSS) measures the perception of stress and thereby how subjects appraise their life as stressful. The 10 item scale is scored on a Likert scale of 0 (never) to 4 (very often) and the items are totalled after reverse scoring of relevant items. The scale has been shown to have good internal consistency in systematic reviews (Cronbach's alpha > 0.7 in majority of studies), as well as good validity and reliability (Lee, 2012). Despite this, this scale showed unacceptable internal consistency in this study ($\alpha = 0.33$). Intercorrelations of items were checked, and three items (4, 5, and 7) were poorly correlated to the total score. However these items were not different in variance from other items. These questions examine confidence about ability to handle problems, feeling like things are going your way and ability to control irritations in your life respectively. It is possible that these questions were less suited to the current population due to the inability to control the child's health or symptoms. While a short form of the scale can be calculates using four questions (2, 4, 5, and

10), the internal consistency of this was still poor ($\alpha = 0.59$). Due to the poor internal consistency in both versions, PSS data was excluded from further analyses within the study.

2.5.8 Quality of Life Scale

The Quality of Life Scale (QoLS) is a 16 item questionnaire that measures the following domains of quality of life: material and physical wellbeing, recreation, relationships with others, social, independence, personal development, and community activities (Burckhardt & Anderson, 2003). The scale has good internal consistency (Cronbach's alpha of 0.82) and high test-retest reliability with 0.78 to 0.84 respectively over 3 weeks (Burckhardt et al.,1989). In the present study this scale demonstrated good internal consistency ($\alpha = 0.87$).

2.6 Power Calculations

An *a priori* power analysis was performed to estimate the required sample size. G*Power analysis showed that, based on a linear multiple regression fixed model *r2* deviation from 0, 3 predictors, a minimum sample size of 77 was required for power of 0.8 with 0.05 significance level. A further calculation using Green's (1991) formula ($N \geq 50+8m$) where m denotes the independent variables showed that a minimum sample size of 66 was required with two independent variables. Cohen (1992) recommended that correlation analyses require a minimum sample of 84 participants to detect a moderate correlation with 80% power and an alpha level of 0.05. As such, this study expected to recruit a minimum 90 participants to allow for the average dropout rate of 30% of participants in online surveys (Galesic, 2006). The final sample size (N=205) was therefore deemed to be sufficiently powered to perform the planned analysis. The survey was designed following principles to minimise dropout such as shortening its length, providing a progress bar and giving an estimated completion time.

2.7 Statistical Analysis

Statistical analyses were conducted using SPSS Version 25 (IBM). Correlations were used to explore hypotheses 1-3 examining the relationships between carer burden, self-compassion, psychological inflexibility and measures of wellbeing (GAD7, PHQ9, and QoLs). Effect sizes were estimated from correlation coefficients and categorised as small medium or large according to Cohen (1988). Pearson's correlations, independent samples t tests, and one way

ANOVAs were used in order to examine the relationship between demographic variables and measures of caregiver wellbeing (GAD7, PHQ9, and QoLS) to determine whether these should be controlled for as confounds in subsequent analyses. In order to examine whether carer burden, self-compassion and psychological inflexibility predict caregiver wellbeing (anxiety, depression and quality of life) stepwise regressions with forward variable selection were computed.

2.7.1 Data screening

Data was screened to ensure that all assumptions necessary for the planned analyses were met. PP plots, histograms and box plots were used to check data assumptions of normality and this was found to be normally distributed. Skewness and kurtosis values were also examined and transformed into *Z* scores to check data normality, and all were within the -1 and +1 range. Linearity and homoscedasticity were checked using scatterplots of residuals, which showed no pattern suggesting that these assumptions were met.

Multicollinearity was checked using Pearson's correlations between independent variables, and all were below 0.8, suggesting that that assumptions were met. The variance inflation factor (VIF) score was also used to assess collinearity and all the values were below 3 indicating that there is no collinearity present in the data. There were no significant outliers present in the data.

2.7.2 Missing Data

There was some missing data identified from the questionnaires, and as no questions were mandatory this was not surprising. Upon further investigation, it was found that the largest missing items with missing data were subjective ratings of job and finance (11.9% and 9.6% respectively) followed by quality of life total scores at 9.6%, carer burden total scores at 8.2%, self-compassion total scores at 8.7%, psychological inflexibility total scores at 3.2%, depression total scores at 2.7%. There was no clear pattern of missing data identified using Little's MCAR test ($X^2= 612.99$, df = (559), $p = 0.56$). Missing data was not imputed due to the fact that most missing data was under 10% for key variables, and in this case pairwise deletion used by statistical software was sufficient to allow for data to be used despite missing cases as suggested by Lodder (2013).

3. Results

3.1 Descriptive Statistics

Descriptive statistics are displayed in Table 3. Scores for the measures indicate that the sample experienced moderate to severe burden, anxiety and depression. It is difficult to examine whether this is representative of other samples due to the variability of demographic characteristics of parents, conditions of children, the measures used across studies in these populations and the accurate reporting of cutoff ranges in other studies. However previous studies examining carer burden in caregivers of children with cancer have found similar mean scores on the ZBI. Wang and colleagues (2017) reported a mean score of 37.7 in parents of children with newly diagnosed leukemia while Al Qadire and colleagues (2020) reported a mean ZBI score of 38.1 in parents of children with cancer. Toledano-Toledano and colleagues (2020) reported a mean score of 23 on the ZBI when examining burden in parents of children with various conditions. With regards to depression scores using the PHQ9, population norm prevalence falls at 5.6% for moderate to severe depressive symptoms (Kocalevent, Hinz & Brahler, 2013) which reinforces that this population experiences higher rates of depressive symptoms (22.9% moderate to severe). In a sample of caregivers of children with chronic conditions, Khanna and colleagues (2015) found 38% to have moderate to severe symptoms using the PHQ9. Population norm prevalence for anxiety measured using the GAD7 was found to be around 5% for moderate anxiety (Lowe et al., 2008), which demonstrates that the current study sample had considerably higher prevalence of anxiety with 30.2% reporting moderate anxiety. Khanna and colleagues (2015) found 17% of their sample to experience moderate to severe anxiety compared to the 21.5% found in the present study. Population norms for quality of life has been reported to be an average score of 90 in healthy individuals (Burckhardt & Anderson, 2003). The present sample reported a lower quality of life at a mean of 68.6, in line with expected lower quality of life for caregivers.

Table 3

Means and Cut off Scores for Measures

Measure	***N***	**Mean (*SD*)**	**Range in sample**
ZBI (carer burden)	189	42.9 (12.5)	14 – 78
AAQ II (psychological inflexibility)	199	23.2 (9.1)	7 – 48
SCS (self-compassion)	186	74.6 (17.2)	33 – 126
QOL (quality of life)	184	68.6 (12.6)	37 – 95

GAD7 (anxiety)	203	9.9 (5.8)	0 – 21
PHQ9 (depression)	199	10.4 (6.1)	0 – 26
Measure cut off scores		***N* (%)**	
Carer Burden			
Little or no burden (0-21)		7 (3.4%)	
Mild to moderate burden (21-40)		70 (34.1%)	
Moderate to severe burden (41-60)		96 (46.8%)	
Severe burden (61-88)		16 (7.8%)	
Anxiety			
Mild anxiety (0-5)		54 (26.3%)	
Moderate anxiety (6-10)		62 (30.2%)	
Moderately severe anxiety (11-15)		44 (21.5%)	
Severe anxiety (16-21)		43 (21%)	
Depression			
Mild depression (0-5)		46 (22.4%)	
Moderate depression (6-10)		61 (29.8%)	
Moderately severe depression (11-15)		47 (22.9%)	
Severe depression (16-20)		51 (24.9%)	

Note: ZBI = Zarit Burden Interview, SCS = Self compassion scale, AAQ II = Acceptance and Action Questionnaire II, QoL = Quality of Life Scale, GAD7 = Generalized Anxiety Disorder Questionnaire 7, PHQ9 = Patient Health Questionnaire 9

3.2 Examining demographic variables as potential confounds for measures of caregiver wellbeing

Correlation analyses were conducted to determine whether there was a relationship between demographic variables and dependent variables (DVs) of anxiety, depression and quality of life, and whether these needed to be included as confounding variables in further analyses. This included examination of both significance and effect sizes (small and medium effect sizes were not included as confounds in further analyses).

Pearson's correlations were computed for scale variables and showed that the number of health conditions was negatively related with quality of life ($r = -0.227$, $p < 0.01$). Subjective ratings of finances were positively correlated with quality of life ($r = 0.211$, $p < 0.01$) and negatively correlated with anxiety ($r = -0.199$, $p < 0.01$) and depression ($r = -0.270$, $p < 0.01$). Perceived job respect was positively correlated with quality of life ($r = 0.205$, $p < 0.01$) and negatively related to depression ($r = -0.153$, $p < 0.05$). There was no significant relationship between any of the DVs and age of child, time since diagnosis, caregiver age, subjective level of education, number of other children or the number of other children with conditions. Effect size was calculated sing Cohen's (1988) cut off scores for r, and none of these were shown to be large effect sizes (greater than 0.5) and so were not included in further analyses.

Independent t- tests showed no significant gender differences for any DVs. Independent sample t-tests showed significant differences for anxiety scores between those with a previous mental health diagnoses (M = 11.1, SD = 6) and those without a previous mental health diagnosis (M = 9.2, SD = 5.64), $t(167.5) = -2.47$, $p < 0.05$, and in depression scores between those with a previous mental health diagnoses (M = 11.7, SD = 6.1) and those without a previous mental health diagnosis (M =9.5, SD = 5.97), $t(167) = - 2.48$, $p < 0.01$). Independent sample t-tests showed significant differences for depression scores between those caregivers with a health condition (M = 11.6, SD = 6) and those without (M = 8.9, SD = 5.9), $t(196) = -3.55$, $p< 0.01$. Cohen's D was calculated for these factors and none were found to have a large effect size (greater than 0.5) and therefore were not included further in analyses.

One Way ANOVAs were carried out to examine the relationship between demographic factors and DVs. Significant differences were found in anxiety scores based on employment status ($F(3, 198) = 4.98$, $p < 0.01$). Results showed significant differences in depression scores based on relationship status ($F(4, 194) = 2.53$, $p = 0.42$), education ($F(3, 195) = 3.83$, $p = 0.01$) and employment status ($F(3, 194) = 5.93$, $p < 0.01$). Results showed significant differences in quality of life scores based on the type of condition ($F(7, 175) = 2.73$, $p = 0.01$), caregiver relationship status ($F(4, 179) = 2.54$, $p = 0.041$), and employment status ($F(3, 179) = 2.94$, $p < 0.05$). Results showed no differences based on country of residence and ethnicity for any variables. Partial eta squared was used to determine effect sizes, and as none of the factors showed large effect sizes (cut off at 0.14 for n^2) these were not included further in analyses.

3.3 Relationships between carer burden, self-compassion, psychological inflexibility and caregiver wellbeing

Pearson's correlations were conducted to examine the connection between carer burden, self-compassion, psychological inflexibility, and caregiver wellbeing (anxiety, depression and quality of life). Results are illustrated in Table 4 below. These findings supported Hypothesis 1-3.

Higher carer burden was associated with lower self-compassion. Correlation coefficients suggested a small effect size (*r* = - 0.25). Higher carer burden was also associated with higher psychological inflexibility. Correlation coefficients suggested a medium effect size (*r* = 0.40). Higher self-compassion was associated with lower psychological inflexibility. Correlation coefficients suggested a large effect size (*r* = - 0.67). Higher carer burden was associated with higher levels of anxiety and depression. Correlation coefficients suggest a medium effect size (*r* = 0.43 and *r* = 0.33 respectively). Higher carer burden was also associated with lower quality of life scores. Correlation coefficients suggest a medium effect size (*r* = -0.46). Higher self-compassion was found to relate to lower levels of anxiety and depression, and correlation coefficients suggest a large effect size (*r* = -0.53 and *r* = - 0.51 respectively). Higher self-compassion was also related to higher quality of life, and correlation coefficients suggest a medium effect size (*r* = 0.41). Higher scores for psychological inflexibility were associated with higher levels of anxiety and depression, and correlation coefficients suggest a large effect size (*r* = 0.66 and *r* = 0.58 respectively). Higher psychological inflexibility was also linked with lower quality of life, correlation coefficients suggest a large effect size (*r* = - 0.56).

Table 4

Correlations Between Carer Burden, Self-compassion, Psychological Inflexibility, Anxiety, Depression and Quality of Life

	ZBI	**SCS**	**AAQ-II**	**QoL**	**GAD7**	**PHQ9**
ZBI	.					
SCS	-0.25**	.				
AAQ – II	0.40**	-0.67**	.			
QoL	- 0.46**	0.41**	-0.56**	.		
GAD7	0.43**	-0.53**	0.66**	-0.48**	.	
PHQ9	0.33**	-0.51**	0.58**	-0.51**	0.72**	.

Note: ** denotes $p < 0.01$ ZBI = Zarit Burden Interview, SCS = Self compassion scale, AAQ II = Acceptance and Action Questionnaire II, QoL = Quality of Life Scale, GAD7 = Generalized Anxiety Disorder Questionnaire 7, PHQ9 = Patient Health Questionnaire 9

3.4 Carer burden, self-compassion and psychological inflexibility as predictors of caregiver wellbeing

Stepwise regression models (forward selection of variables) were computed to examine whether carer burden, self-compassion and psychological inflexibility predict caregiver wellbeing measures of anxiety, depression and quality of life. Findings supported hypothesis 4, with the exception of the finding that carer burden did not significantly predict depression as anticipated. Effect sizes for the total model regression were calculated using Cohens f^2(1988) which suggests that an $f^2 \geq 0.02$ represents small, ≥ 0.15 moderate and ≥ 0.35 large effect sizes. Effect sizes for individual predictors using β were estimated based on Acock (2014) who suggested that $\beta < 0.2$ is considered a weak effect, 0.2 to <0.50 moderate, and >0.5 a strong effect.

3.4.1 Anxiety

A stepwise regression was conducted with anxiety as the dependent variable and carer burden, self-compassion and psychological inflexibility as the independent variables. The predictors accounted for 45% of variance. It was found that combined carer burden, self-compassion and psychological inflexibiltiy explain a significant proportion of variance in caregiver anxiety ($F(3,168) = 47.95$, $p = 0.00$, $R^2 = 0.46$, $R^2_{Adjusted} = 0.45$). The effect size using Cohen's f^2 was found to be 0.85 indicating a large effect size. Carer burden ($\beta = 0.10$, t(171), = 3.73, p = 0.00) and self-compassion ($\beta = -0.06$, t(171), =,-2.57 p = 0.01) significantly predicted anxiety in caregivers with small effect sizes. Psychological inflexibility ($\beta = 0.26$, t(171), = 5.08, p = 0.00) significantly predicted anxiety in caregivers with a moderate effect size.

3.4.2 Depression

A stepwise regression was conducted with depression as the dependent variable and carer burden, self-compassion and psychological inflexibility as the independent variables. The predictors accounted for 32% of variance. It was found that in combination carer burden, self-compassion and psychological inflexibiltiy explain a significant proportion of variance in caregiver depression ($F(3,164) = 27.29$, $p = 0.00$, $R^2 = 0.33$, $R^2_{Adjusted} = 0.32$). The effect size using Cohen's f^2 was found to be 0.49 indicating a large effect size. Carer burden ($\beta = 0.06$, $t(167) = 1.96$, $p = 0.52$) did not significantly predict depression, and showed a weak effect size. Self-compassion (β = -0.08 , t(167) = - 0.25, p = 0.004) significantly predicted depression in caregivers with a weak effect size. Psychological inflexibility (β = 0.21, t(167) = 3.48, p = 0.001) significantly predicted depression in caregivers with a moderate effect size.

3.4.3 Quality of Life

A stepwise regression was conducted with quality of life as the dependent variable and carer burden, self-compassion and psychological inflexibility as the independent variables. The predictors accounted for 38% of variance. It was found that the combination of carer burden, self-compassion and psychological inflexibiltiy explain a significant proportion of variance in caregiver quality of life ($F(3,156)$ = 32.998, p = 0.00, R^2 = 0.39, $R^2_{Adjusted}$ = 0.38). The effect size using Cohen's f^2 was found to be 0.64 indicating a large effect size. Carer burden (β = -0.32, t(159) = -4.79, p = 0.00) significantly predicted quaity of life with a moderate effect size. Self-compassion (β = 0.13, t(159) = 0.18, p = 0.03) significantly predicted quality of life with a weak effect size and psychological inflexibility (β = -0.396 , t(159) = -3.36, p = 0.00) significantly predicted quality of life in caregivers with a moderate effect size.

4. Discussion

This study examined the relationships between carer burden, self-compassion, psychological inflexibility and caregiver well-being in caregivers of children with chronic conditions.

4.1 Carer Burden

As anticipated, higher levels of carer burden were correlated with higher levels of depression anxiety and lower levels of quality of life in caregivers in the present sample (medium effect sizes). Regression analyses confirmed that higher levels of carer burden predicted higher levels of anxiety and depression and lower levels of quality of life (large effect size). Similar findings were found in prior studies examining links between carer burden and mental health (Santo et al., 2011), depression and anxiety (Gallagher et al., 2008) and quality of life (Collins et al., 2020). This further highlights the importance of addressing carer burden in this population due to the impact on caregiver wellbeing.

Higher carer burden was also found to be correlated with lower self-compassion (small effect size). This is in line with previous findings showing a relationship between carer burden with self-compassion (Bohadana et al., 2019, Neff & Faso, 2015). Higher carer burden was also correlated with higher psychological inflexibility (medium effect size). This was similar to previous research findings of a medium effect size in the relationship between burden and psychological inflexibility (Slowey, 2014). This suggests a need for further examination of the role of self-compassion and psychological inflexibility in affecting burden in caregivers of children with chronic conditions

4.2 Self-compassion

Results also emphasized the importance of self-compassion in this population. Higher self-compassion was found to be correlated to lower levels of anxiety and depression (large effect size) and better quality of life (medium effect size). Regression analyses confirmed that higher levels of self-compassion predicted lower levels of carer burden, depression and anxiety and higher levels of quality of life in caregivers (large effect sizes). The results were similar to findings from previous research evidencing links between self-compassion and mental wellbeing across age groups (MacBeth et al., Marsh et al., 2018) and the relationship with carer burden (Hsieh et al., 2012; Lloyd et al., 2019; Neff & Faso, 2015). The present study further suggests that self-compassion plays a key role in the experience of carer burden and caregiver wellbeing and highlights the importance of supporting caregivers to develop self-compassion in their caring roles.

However, the current study found no evidence that self-compassion moderated the relationship between carer burden and caregiver well-being measures. These findings were inconsistent with previous research into carer burden in caregivers of individuals with lung cancer by Hsieh and colleagues (2019). There are a number of potential explanations for the disparity in findings. Firstly, the study by Hsieh and colleagues (2019) used different measures namely the Kingston Caregivers Stress Scale (Sadak et al., 2017), the 13 Item Self-Compassion Scale (Gilbert et al., 2017) and the 21 Item Beck Depression Inventory (Beck et al., 1996). It is therefore not possible to directly compare the means of their sample to the participants in this study due to variation in scale cut-off scores. Additionally, the sample in the Hsieh study (2019) had a different gender distribution with almost half of caregivers being male. This is unusual in studies of caregivers of children with chronic conditions where most studies have a predominantly female sample. Small gender differences in self-compassion have been found in a meta-analysis (Yarnell et al., 2015), and Hsieh and colleagues did not examine gender as a covariate in their moderation analysis despite their split sample. While these effect sizes are small it could be useful to further examine what effect gender has on the relationship between carer burden, self-compassion and caregiver wellbeing.

4.3 Psychological Flexibility

Results showed a connection between higher psychological inflexibility scores and higher levels of anxiety and depression and lower quality of life (large effect sizes). Regression analyses confirmed that psychological inflexibility predicted higher levels of carer burden, depression, anxiety and lower levels of quality of life in caregivers. This finding is in accordance with previous research by Sairanen and colleagues (2018) which showed that lower levels of psychological flexibility predicted anxiety and depression in caregivers of children with chronic conditions. Psychological flexibility may be a crucial factor for caregivers as they have to adapt to the ever-altering demand of their caring role and the life changes this demands.

While intervention studies seemed to suggest a role for psychological flexibility in acting as a mechanism for improving caregiver wellbeing (Blackledge & Hayes, 2006; Poddar et al., 2015) the present study found no evidence that psychological inflexibility moderated the relationship between carer burden and caregiver well-being measures. These findings are similar to those from a previous study by Slowey (2014) which found that psychological flexibility did not moderate the relationship between burden and wellbeing in informal carers. While the sample from Slowly (2014) was underpowered, the current sample had sufficient power for moderation analyses which suggests that findings are accurate. However, the present studies examined caregivers of children with a variety of conditions (physical, mental, neurodevelopmental). Future research may wish to examine whether there are differences in psychological flexibility between these distinct groups of conditions and compare these to a control group in order to examine whether these results may have been affected by using a mixed sample.

4.4 Self-compassion and psychological inflexibility

As anticipated higher self-compassion was significantly related to lower psychological inflexibility (large effect size). Previous research demonstrated a relationship between these factors and showed their relationship with emotional wellbeing (Marshall & Brockman, 2016). In the present study these factors were moderately correlated, and as such were not found to have high multicollinearity. Future research may wish to further examine the interaction between these factors in this population in order to examine the common mechanisms through which they may be related. This will aid with intervention development and could suggest different models of testing their effects on the relationship between carer burden and caregiver wellbeing.

4.5 Clinical Implications

This study has a number of potential clinical implications. It is clear that caregivers of children with a range of chronic conditions experience high levels of carer burden, high levels of distress (anxiety, depression) and lowered quality of life. This is in line with prior research (Pinquart, 2018). It is therefore important to support caregivers in their role in order to minimise the negative impacts of the caregiving experience on their wellbeing. This research provides preliminary understanding that self-compassion and psychological flexibility are related to improved psychological wellbeing in this population. Therefore, this research emphasizes the importance of considering these factors in a clinical setting, such as early intervention or identification of particular vulnerable carers.

Interventions focusing on increasing self-compassion and psychological flexibility in caregivers may prove beneficial, such as CFT and ACT. However further research is needed in order to better understand the ways in which these factors affect the relationship between burden and wellbeing. In this population in particular, it may be beneficial to consider the development of guided self-help or blended therapy approaches due to the barriers of engaging in a traditional face to face intervention such as the time and resource burden of the caregiving role (Barlow et al., 2006). E-interventions have been shown to reduce difficulties with accessibility due to their flexibility and have been well received by caregivers of children with chronic conditions (Palermo et al., 2009). It is important to further research this area before interventions are developed, as many carer interventions are created before sufficient research is conducted into their efficacy and the mechanisms of change therein (Parker et al., 2010). It is also important to involve caregivers in the design and development of interventions, as often the needs of the population are different to the anticipated needs from professionals creating intervention tools. The Department of Health policy in the UK has highlighted the need for clinicians to involve service users in health intervention development and research for some time (Boote et al., 2002).

4.6 Strengths and Limitations

This study recruited an opportunity sample of volunteers from social media platforms where specific support groups and charity organisations were asked to share the weblink to the survey. By using this recruitment method, the study was able to recruit caregivers from a wide range of locations and whose children had a range of chronic conditions, which has previously been a limitation of studies examining carer burden in this population and improves the generalizability of findings. By adopting a regression approach the present study was able to pinpoint predictors of mental health difficulties in this population, thereby initiating the search for intervention targets. Additionally, the study used a validated tool for measuring carer burden (ZBI) which allows for easier comparison of findings between studies in future reviews and links with other research findings examining carer burden with this tool.

However, findings from the present study should be interpreted in light of a number of limitations. Firstly, the study used a cross-sectional design and so it was unable to determine causal relationships. It will remain difficult to make causal claims in research such as this due to the difficulties of manipulating the factors examined experimentally. Future research might therefore attempt to examine these factors longitudinally in order to examine predictability over time and replicate findings. The recruitment strategy was likely to have resulted in selection bias and thus lower external validity. As such future research may consider recruiting participants from clinical services directly. The sample also consisted of primarily female caregivers, similarly to samples in previous studies of carer burden in this population. The majority of studies seem to consist of mainly female caregivers, and it would be useful to examine whether there is a difference in these factors in male caregivers. This study also did not examine hours of care or whether the caregiver lived with the child (although the second could be assumed due to the age of most children). Future research should incorporate these factors into demographic questionnaires.

Additionally, the Perceived Stress Scale showed poor internal consistency in the present study both with the 10-item version and the 4-item short version, despite the fact that previous studies have used the 14-item scale successfully with caregivers of children with chronic conditions such as type 1 diabetes (Tisouli et al., 2014) and found no difficulties with internal consistency. Potential reasons for poor internal consistency were examined including evaluating the percentage of missing data, intercorrelations, variance and outliers, and potential errors in data entry or question formulation. None of these avenues highlighted a notable issue with the data set. As such the decision was made to exclude this from subsequent analysis. It was felt that removing a number of items which were not correlated with the total score would result in loss of specificity in the scale. The items which were not correlated with the total score related to a sense of controllability, things going your way and ability to control irritations. It is possible that these questions are particularly difficult in a population where the perceived stress is likely related to the child's health which is uncontrollable. Future studies may wish to examine whether other measures of perceived stress, or adaptations of this scale, may be better suited for use in this population.

4.7 Future Research

Future research may consider examining conceptual or theoretical models of carer burden, self-compassion and psychological flexibility in order to focus on specific aspects of this triad and the interaction with demographic variables. Future research may also wish to examine whether caregiver health status influences caregiving experience. Half of the sample had a health condition themselves, and it might be useful to examine whether the type of health condition of the caregiver influences their experience of carer burden (i.e. if this condition is the same or different from the condition experienced by their child). While efforts were made to recruit a sample of caregivers of children with a variety of condition types, majority of the present sample had a physical health condition. Future research may wish to examine carer burden, self-compassion and psychological flexibility in samples consisting of purely mental or neurodevelopmental conditions, and to compare these groups. Most caregivers in this sample experienced moderate or severe burden, one fifth of caregivers reported severe anxiety symptoms and a quarter reported severe depressive symptoms.

Further research is needed in order to better understand the roles of self-compassion and psychological flexibility in this sample to aid in the development of interventions to support caregivers who are evidently experiencing high levels of carer burden and lowered wellbeing.

4.8 Conclusion

This study presents evidence of the associations between carer burden, self-compassion and psychological flexibility with caregiver wellbeing. However it did not show evidence of a moderating effect of self-compassion and psychological flexibility on the effect of carer burden on anxiety, depression or quality of life. This does not necessarily suggest that no such effect exists, but that there is no evidence of this in the present sample. Further research is warranted to examine whether these findings can be replicated across samples, and to continue to examine the potential ways in which self-compassion and psychological flexibility may interplay to impact on caregiver wellbeing.

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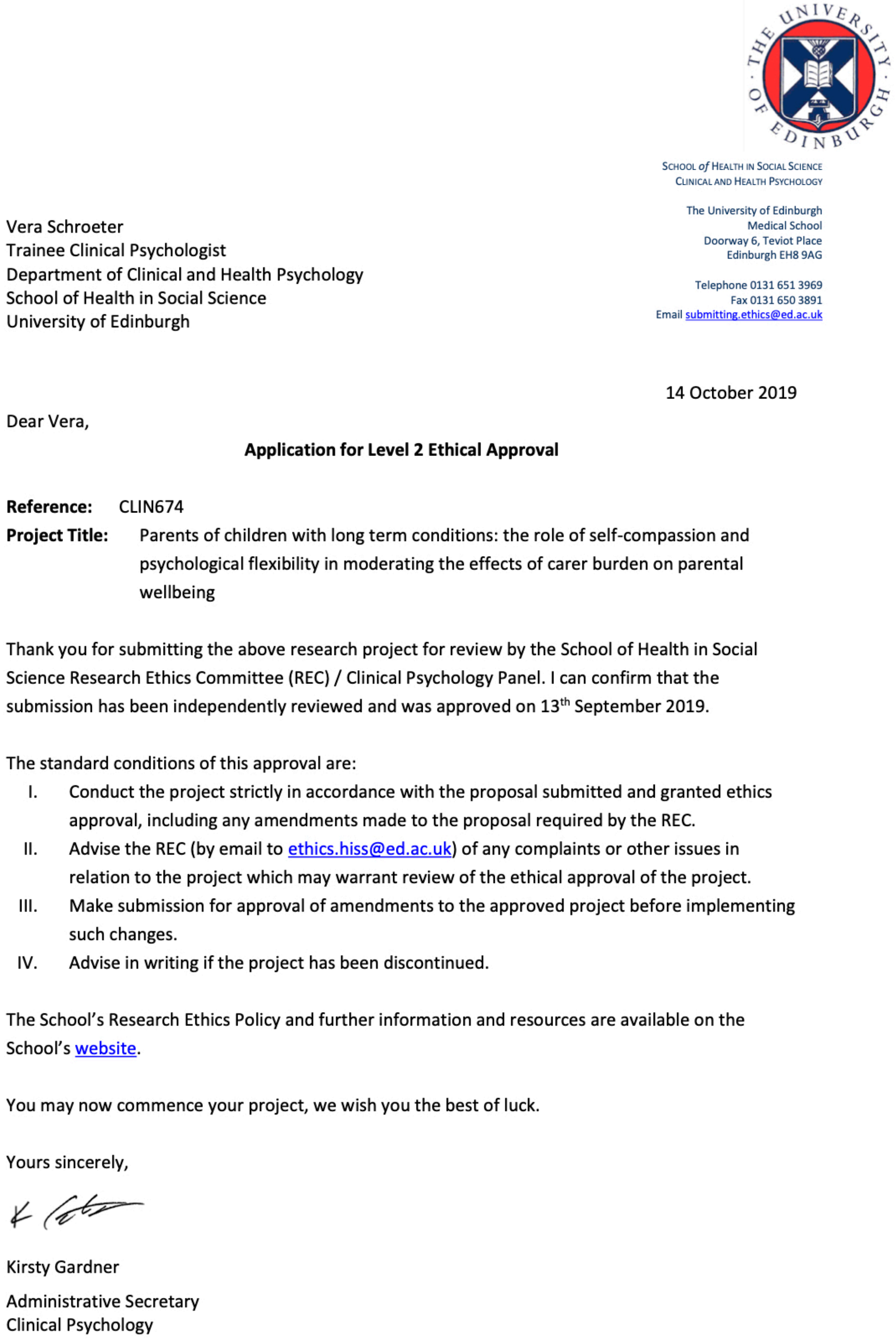
Appendix A: Confirmation of Ethical Approval and Sponsorship



SCHOOL *of* HEALTH IN SOCIAL SCIENCE
CLINICAL AND HEALTH PSYCHOLOGY

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Edinburgh EH8 9AG

Telephone 0131 651 3969
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Email submitting.ethics@ed.ac.uk

Vera Schroeter
Trainee Clinical Psychologist
Department of Clinical and Health Psychology
School of Health in Social Science
University of Edinburgh

14 October 2019

Dear Vera,

Application for Level 2 Ethical Approval

Reference: CLIN674
Project Title: Parents of children with long term conditions: the role of self-compassion and psychological flexibility in moderating the effects of carer burden on parental wellbeing

Thank you for submitting the above research project for review by the School of Health in Social Science Research Ethics Committee (REC) / Clinical Psychology Panel. I can confirm that the submission has been independently reviewed and was approved on 13th September 2019.

The standard conditions of this approval are:

I. Conduct the project strictly in accordance with the proposal submitted and granted ethics approval, including any amendments made to the proposal required by the REC.
II. Advise the REC (by email to ethics.hiss@ed.ac.uk) of any complaints or other issues in relation to the project which may warrant review of the ethical approval of the project.
III. Make submission for approval of amendments to the approved project before implementing such changes.
IV. Advise in writing if the project has been discontinued.

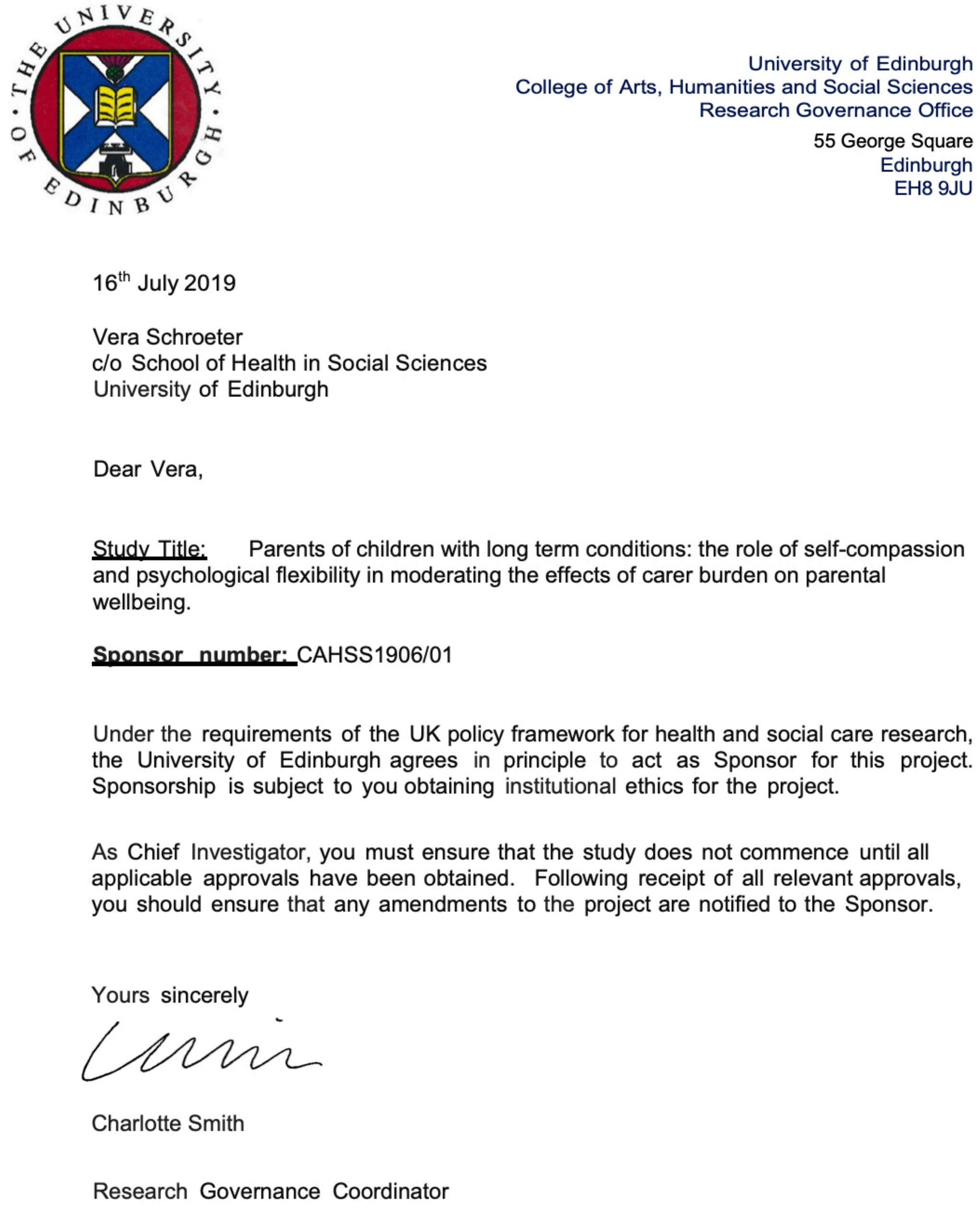
The School's Research Ethics Policy and further information and resources are available on the School's website.

You may now commence your project, we wish you the best of luck.

Yours sincerely,

Kirsty Gardner

Administrative Secretary
Clinical Psychology



University of Edinburgh
College of Arts, Humanities and Social Sciences
Research Governance Office
55 George Square
Edinburgh
EH8 9JU

16th July 2019

Vera Schroeter
c/o School of Health in Social Sciences
University of Edinburgh

Dear Vera,

Study Title: Parents of children with long term conditions: the role of self-compassion and psychological flexibility in moderating the effects of carer burden on parental wellbeing.

Sponsor number: CAHSS1906/01

Under the requirements of the UK policy framework for health and social care research, the University of Edinburgh agrees in principle to act as Sponsor for this project. Sponsorship is subject to you obtaining institutional ethics for the project.

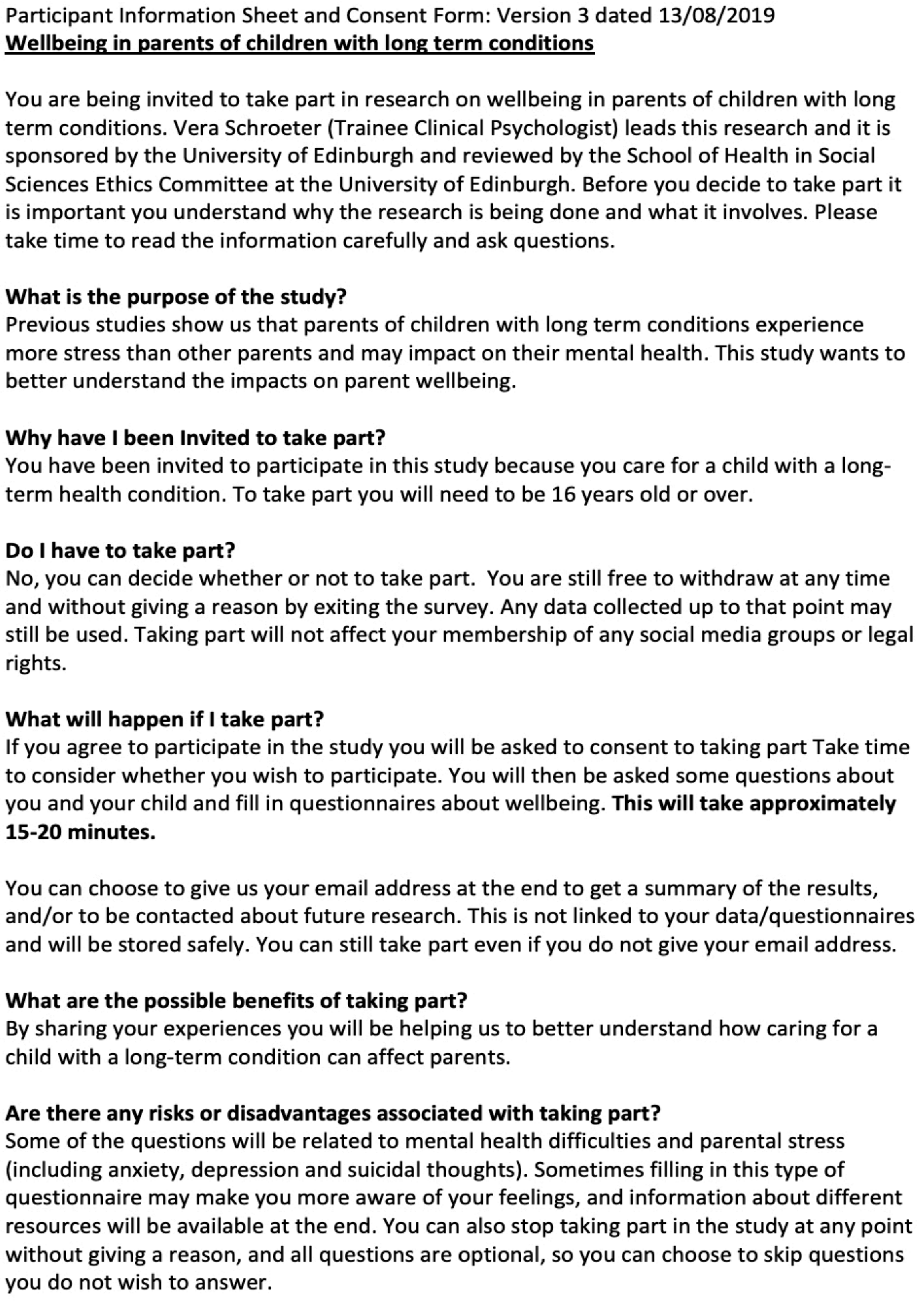
As Chief Investigator, you must ensure that the study does not commence until all applicable approvals have been obtained. Following receipt of all relevant approvals, you should ensure that any amendments to the project are notified to the Sponsor.

Yours sincerely

Charlotte Smith

Research Governance Coordinator

Appendix B: Information and Consent Forms.



Participant Information Sheet and Consent Form: Version 3 dated 13/08/2019

Wellbeing in parents of children with long term conditions

You are being invited to take part in research on wellbeing in parents of children with long term conditions. Vera Schroeter (Trainee Clinical Psychologist) leads this research and it is sponsored by the University of Edinburgh and reviewed by the School of Health in Social Sciences Ethics Committee at the University of Edinburgh. Before you decide to take part it is important you understand why the research is being done and what it involves. Please take time to read the information carefully and ask questions.

What is the purpose of the study?

Previous studies show us that parents of children with long term conditions experience more stress than other parents and may impact on their mental health. This study wants to better understand the impacts on parent wellbeing.

Why have I been Invited to take part?

You have been invited to participate in this study because you care for a child with a long-term health condition. To take part you will need to be 16 years old or over.

Do I have to take part?

No, you can decide whether or not to take part. You are still free to withdraw at any time and without giving a reason by exiting the survey. Any data collected up to that point may still be used. Taking part will not affect your membership of any social media groups or legal rights.

What will happen if I take part?

If you agree to participate in the study you will be asked to consent to taking part Take time to consider whether you wish to participate. You will then be asked some questions about you and your child and fill in questionnaires about wellbeing. **This will take approximately 15-20 minutes.**

You can choose to give us your email address at the end to get a summary of the results, and/or to be contacted about future research. This is not linked to your data/questionnaires and will be stored safely. You can still take part even if you do not give your email address.

What are the possible benefits of taking part?

By sharing your experiences you will be helping us to better understand how caring for a child with a long-term condition can affect parents.

Are there any risks or disadvantages associated with taking part?

Some of the questions will be related to mental health difficulties and parental stress (including anxiety, depression and suicidal thoughts). Sometimes filling in this type of questionnaire may make you more aware of your feelings, and information about different resources will be available at the end. You can also stop taking part in the study at any point without giving a reason, and all questions are optional, so you can choose to skip questions you do not wish to answer.

Will my taking part in the study be kept confidential?
The University of Edinburgh (data controller) sponsors this study and is responsible for looking after your information. Identifiable information about you will be kept for ten years and anonymous data indefinitely. We will use the minimum personally-identifiable information possible. You have limited rights to change your data as we manage the information in a specific way for accuracy. If you stop the study we will keep the collected information about you.

Your data will be processed in accordance with Data Protection Law. All information collected about you will be kept strictly confidential and will only be viewed by the researcher/research team. University of Edinburgh generic privacy notice: https://www.ed.ac.uk/records-management/privacy-notice-research

What happens when the study is finished?
At the end of the research we will keep unidentifiable data without your email address or personal details in a secure location indefinitely. If you consented to be contacted with a summary of the research, you will be sent this via email at the end of the study.

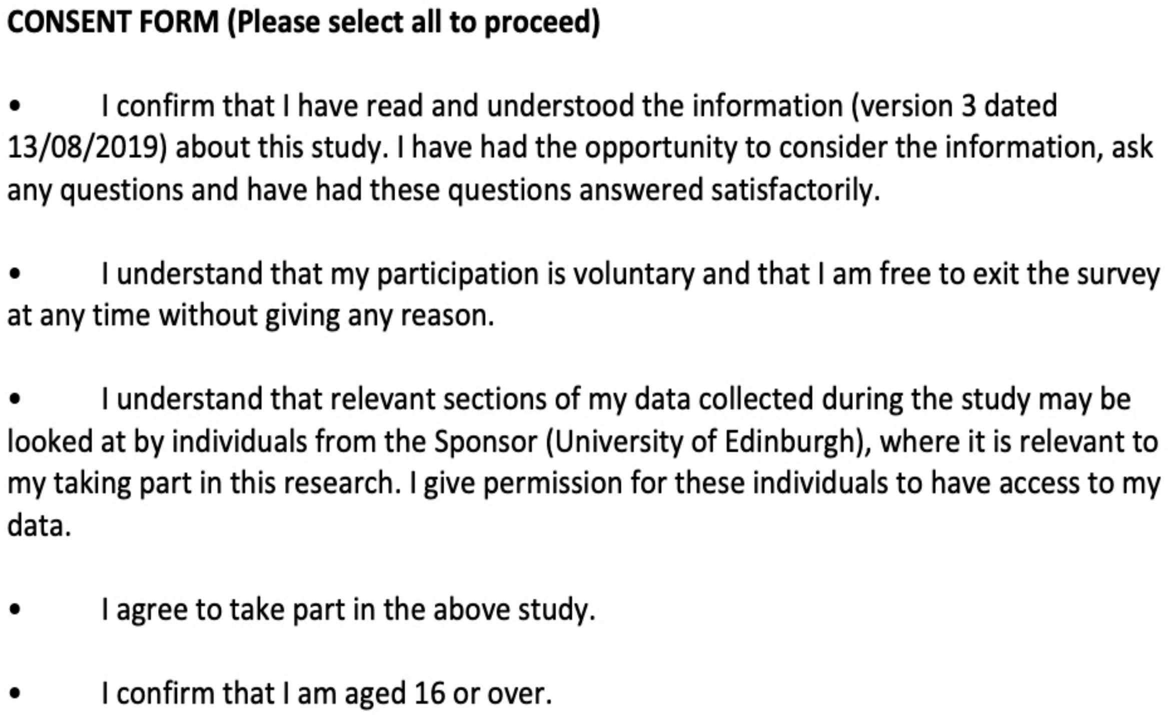
What will happen to the results of the study?
The study will be written up as a thesis and published in relevant journals in due time. You will not be identifiable in any published results.

Who has reviewed the study?
The study proposal has been reviewed by the School of Health in Social Sciences Ethics Committee at the University of Edinburgh.

WHO CAN I CONTACT?
If you have any further questions about the study, please contact the lead researcher Vera Schroeter, vera.schroeter@nhs.net. To contact her supervisor Dr Stella Chan please email stella.chan@ed.ac.uk. If you would like to discuss this study with someone independent of the study please contact Dr Ethel Quayle, ethel.quayle@ed.ac.uk.

What if there is a problem?
If you have a concern about any aspect of this study please contact Vera Schroeter (vera.schroeter@nhs.net) who will do her best to answer your questions.

If you wish to make a complaint about the study, please contact: The University of Edinburgh Research Governance team at cahss.res.ethics@ed.ac.uk



CONSENT FORM (Please select all to proceed)

- I confirm that I have read and understood the information (version 3 dated 13/08/2019) about this study. I have had the opportunity to consider the information, ask any questions and have had these questions answered satisfactorily.

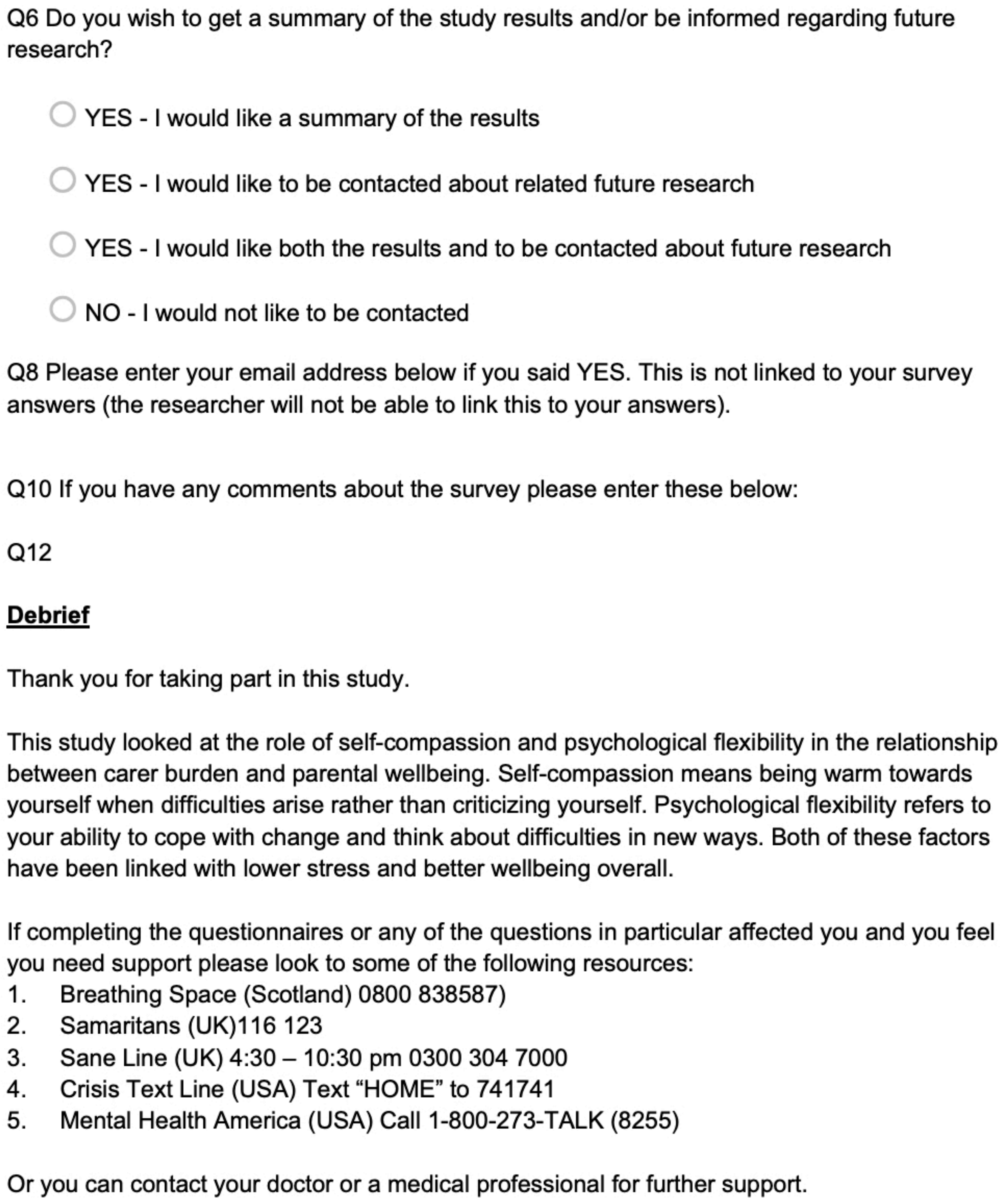
- I understand that my participation is voluntary and that I am free to exit the survey at any time without giving any reason.

- I understand that relevant sections of my data collected during the study may be looked at by individuals from the Sponsor (University of Edinburgh), where it is relevant to my taking part in this research. I give permission for these individuals to have access to my data.

- I agree to take part in the above study.

- I confirm that I am aged 16 or over.

Appendix C: Debrief Form.



Q6 Do you wish to get a summary of the study results and/or be informed regarding future research?

○ YES - I would like a summary of the results

○ YES - I would like to be contacted about related future research

○ YES - I would like both the results and to be contacted about future research

○ NO - I would not like to be contacted

Q8 Please enter your email address below if you said YES. This is not linked to your survey answers (the researcher will not be able to link this to your answers).

Q10 If you have any comments about the survey please enter these below:

Q12

Debrief

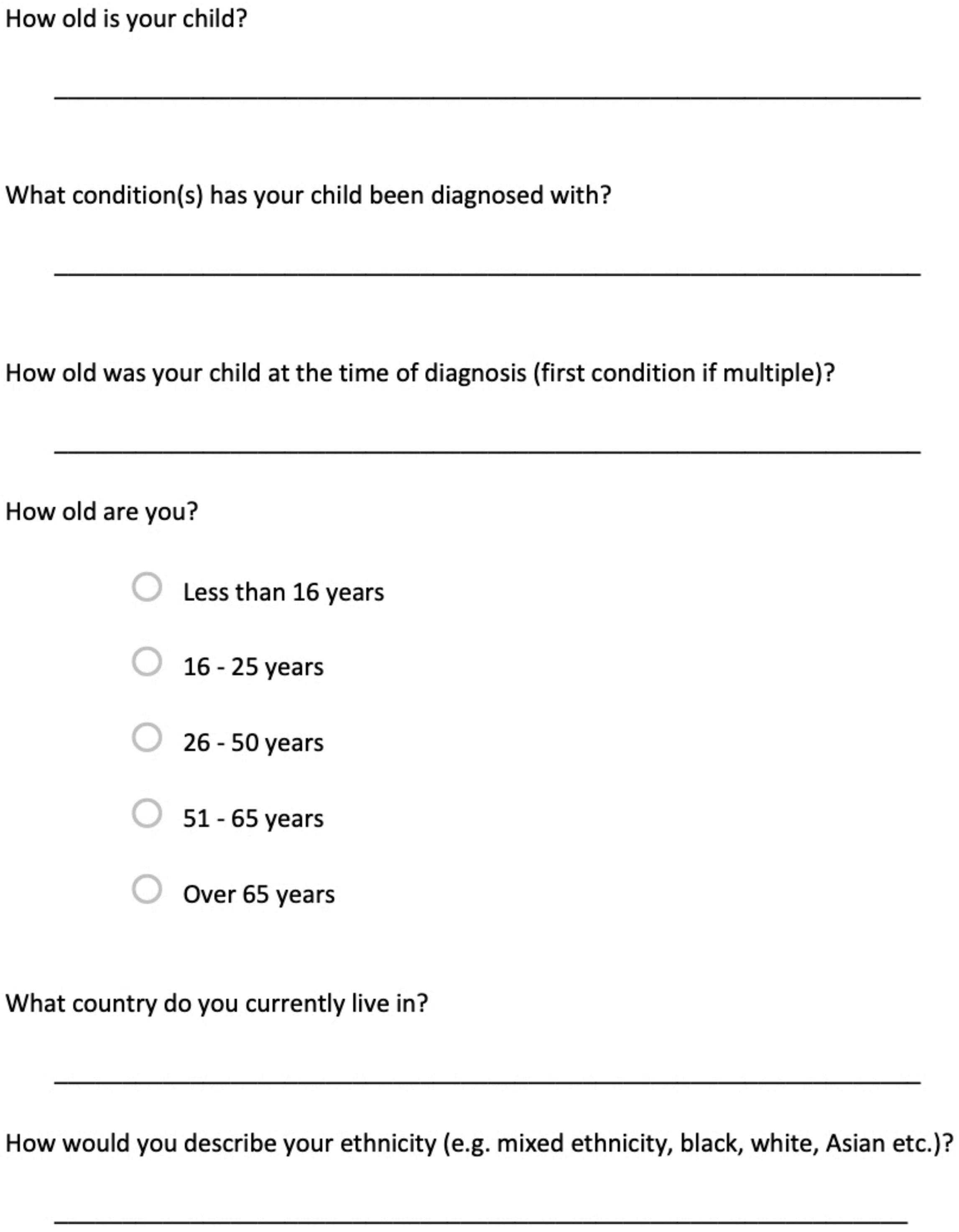
Thank you for taking part in this study.

This study looked at the role of self-compassion and psychological flexibility in the relationship between carer burden and parental wellbeing. Self-compassion means being warm towards yourself when difficulties arise rather than criticizing yourself. Psychological flexibility refers to your ability to cope with change and think about difficulties in new ways. Both of these factors have been linked with lower stress and better wellbeing overall.

If completing the questionnaires or any of the questions in particular affected you and you feel you need support please look to some of the following resources:
1. Breathing Space (Scotland) 0800 838587)
2. Samaritans (UK)116 123
3. Sane Line (UK) 4:30 – 10:30 pm 0300 304 7000
4. Crisis Text Line (USA) Text “HOME” to 741741
5. Mental Health America (USA) Call 1-800-273-TALK (8255)

Or you can contact your doctor or a medical professional for further support.

Appendix D. Demographic Questionnaires.



How old is your child?

____

What condition(s) has your child been diagnosed with?

____

How old was your child at the time of diagnosis (first condition if multiple)?

____

How old are you?

○ Less than 16 years

○ 16 - 25 years

○ 26 - 50 years

○ 51 - 65 years

○ Over 65 years

What country do you currently live in?

____

How would you describe your ethnicity (e.g. mixed ethnicity, black, white, Asian etc.)?

____

What is your relationship status?

- ☐ Single
- ☐ In a relationship
- ☐ Married
- ☐ Widowed
- ☐ Separated/Divorced
- ☐ Other ____

What is the highest level of education you have completed?

- ○ 1 = university or college or equivalent
- ○ 2 = intermediate between secondary level and university (e.g. technical training)
- ○ 3 = secondary school
- ○ 4 = primary school only (or less)

Where would you rate yourself compared to others in your country (slide to indicate) 0 = least and 100=most

	0 10 20 30 40 50 60 70 80 90 100
The amount of money you have (1)	
The amount of education you have (2)	
How respected your job is (3)	

What is your current job status?

- ○ Unemployed
- ○ Part-time
- ○ Full time
- ○ Other ____

How would you describe where you live?

- ○ In a town/city
- ○ In the suburbs (near a town/city)
- ○ In the countryside

How would you describe your gender?

____

How many other children do you have?

- ○ 0
- ○ 1
- ○ 2
- ○ 3
- ○ 4
- ○ 5
- ○ 6
- ○ 7
- ○ 8
- ○ 9
- ○ 10+

How many of your other children also have a long-term health condition?

○ 0

○ 1

○ 2

○ 3

○ 4

○ 5

○ 6

○ 7

○ 8

○ 9

○ 10+

Did you have any mental health difficulties before your child was diagnosed?

○ No

○ Yes (please specify) ____

Do you have any health conditions yourself?

○ No

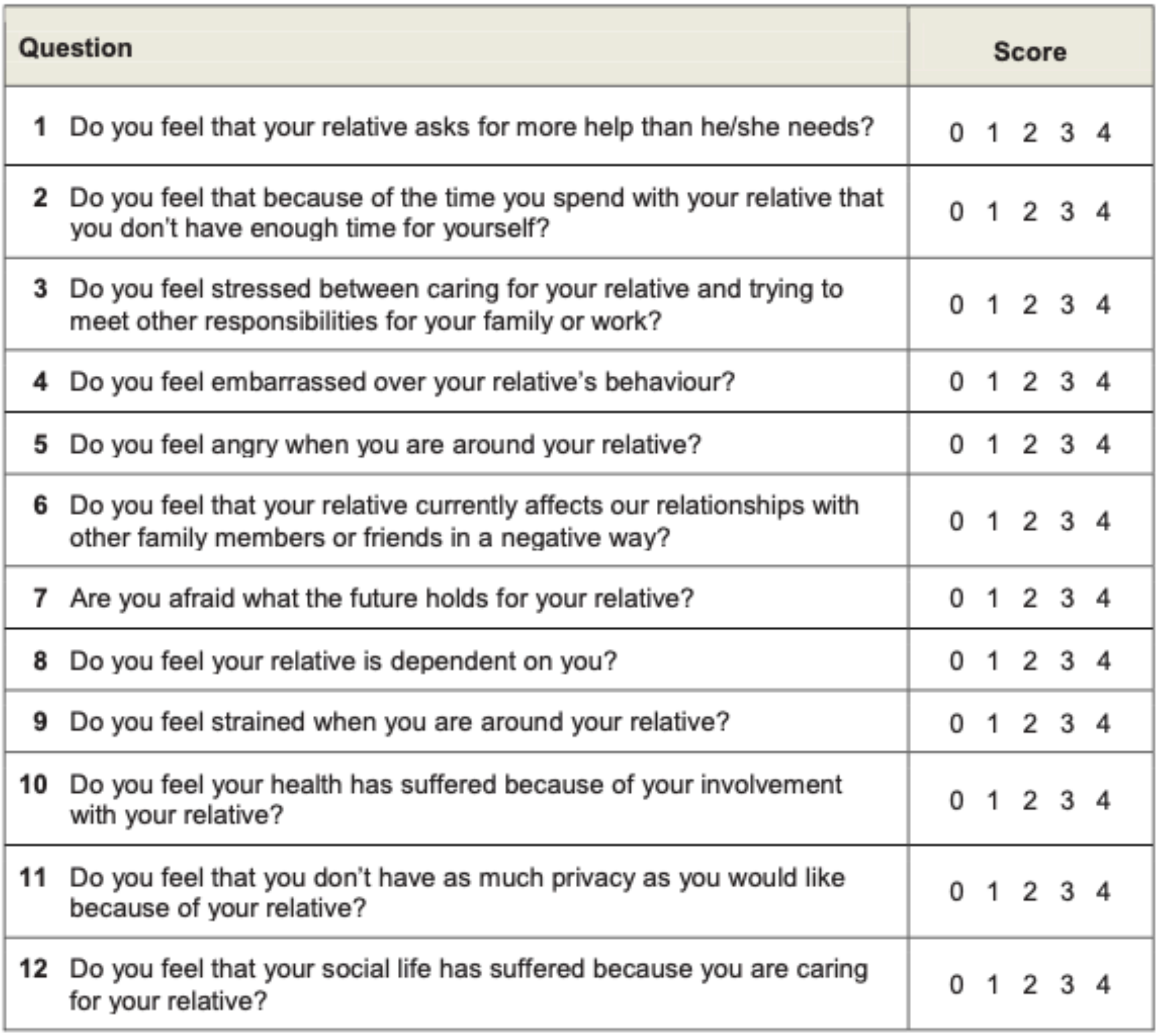
○ Yes (please specify) ____

Appendix E: Measures and Questionnaires

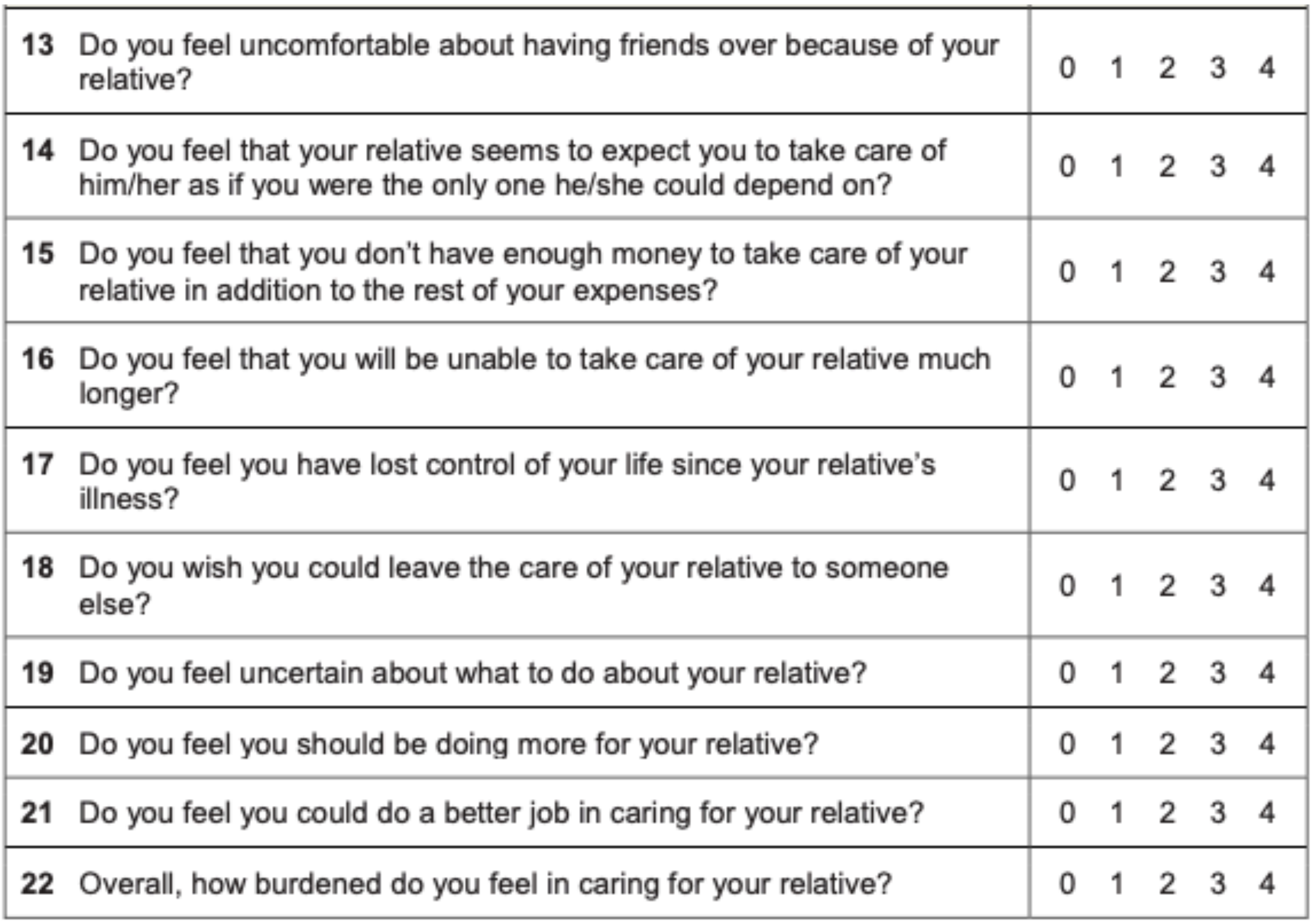
Zarit Burden Interview

Please circle the response the best describes how you feel.

0: never 1: rarely 2: sometimes 3: quite frequently 4: nearly always

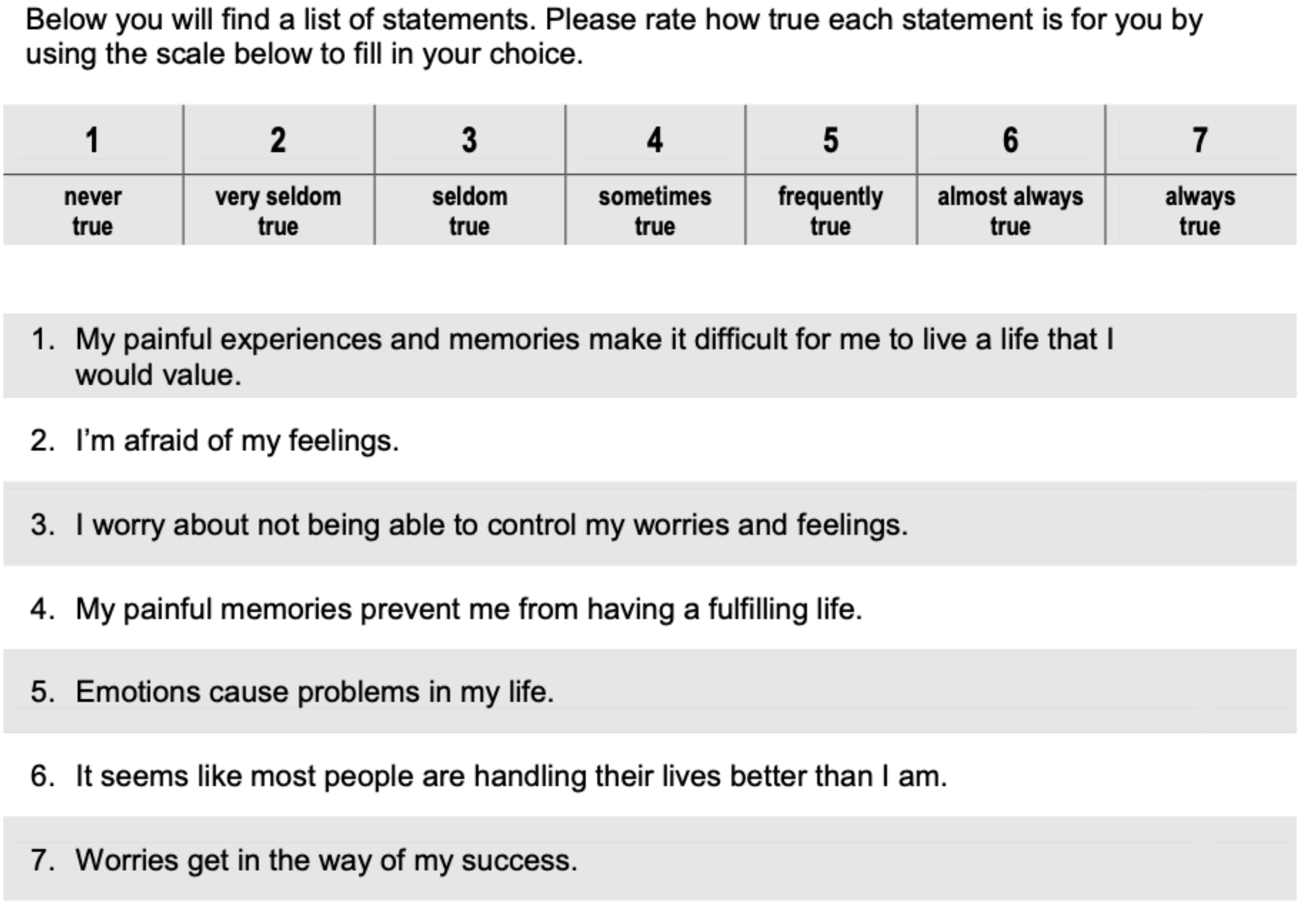


	Question	Score
1	Do you feel that your relative asks for more help than he/she needs?	0 1 2 3 4
2	Do you feel that because of the time you spend with your relative that you don't have enough time for yourself?	0 1 2 3 4
3	Do you feel stressed between caring for your relative and trying to meet other responsibilities for your family or work?	0 1 2 3 4
4	Do you feel embarrassed over your relative's behaviour?	0 1 2 3 4
5	Do you feel angry when you are around your relative?	0 1 2 3 4
6	Do you feel that your relative currently affects our relationships with other family members or friends in a negative way?	0 1 2 3 4
7	Are you afraid what the future holds for your relative?	0 1 2 3 4
8	Do you feel your relative is dependent on you?	0 1 2 3 4
9	Do you feel strained when you are around your relative?	0 1 2 3 4
10	Do you feel your health has suffered because of your involvement with your relative?	0 1 2 3 4
11	Do you feel that you don't have as much privacy as you would like because of your relative?	0 1 2 3 4
12	Do you feel that your social life has suffered because you are caring for your relative?	0 1 2 3 4



13	Do you feel uncomfortable about having friends over because of your relative?	0	1	2	3	4
14	Do you feel that your relative seems to expect you to take care of him/her as if you were the only one he/she could depend on?	0	1	2	3	4
15	Do you feel that you don't have enough money to take care of your relative in addition to the rest of your expenses?	0	1	2	3	4
16	Do you feel that you will be unable to take care of your relative much longer?	0	1	2	3	4
17	Do you feel you have lost control of your life since your relative's illness?	0	1	2	3	4
18	Do you wish you could leave the care of your relative to someone else?	0	1	2	3	4
19	Do you feel uncertain about what to do about your relative?	0	1	2	3	4
20	Do you feel you should be doing more for your relative?	0	1	2	3	4
21	Do you feel you could do a better job in caring for your relative?	0	1	2	3	4
22	Overall, how burdened do you feel in caring for your relative?	0	1	2	3	4

AAQ II Questionnaire

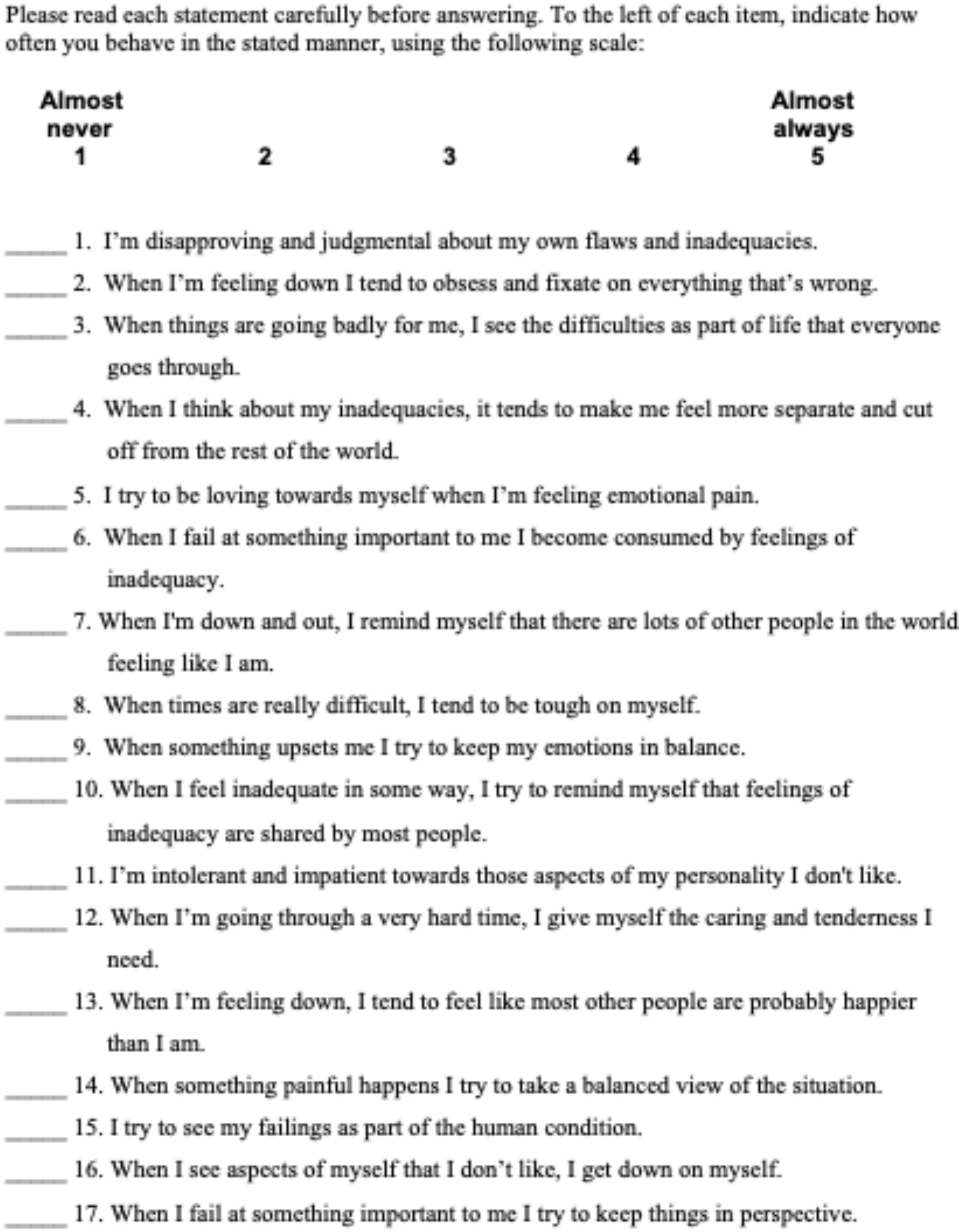


Below you will find a list of statements. Please rate how true each statement is for you by using the scale below to fill in your choice.

1	2	3	4	5	6	7
never true	**very seldom true**	**seldom true**	**sometimes true**	**frequently true**	**almost always true**	**always true**

1. My painful experiences and memories make it difficult for me to live a life that I would value.
2. I'm afraid of my feelings.
3. I worry about not being able to control my worries and feelings.
4. My painful memories prevent me from having a fulfilling life.
5. Emotions cause problems in my life.
6. It seems like most people are handling their lives better than I am.
7. Worries get in the way of my success.

Self-Compassion Scale



Please read each statement carefully before answering. To the left of each item, indicate how often you behave in the stated manner, using the following scale:

Almost never				Almost always
1	2	3	4	5

_____ 1. I'm disapproving and judgmental about my own flaws and inadequacies.
_____ 2. When I'm feeling down I tend to obsess and fixate on everything that's wrong.
_____ 3. When things are going badly for me, I see the difficulties as part of life that everyone goes through.
_____ 4. When I think about my inadequacies, it tends to make me feel more separate and cut off from the rest of the world.
_____ 5. I try to be loving towards myself when I'm feeling emotional pain.
_____ 6. When I fail at something important to me I become consumed by feelings of inadequacy.
_____ 7. When I'm down and out, I remind myself that there are lots of other people in the world feeling like I am.
_____ 8. When times are really difficult, I tend to be tough on myself.
_____ 9. When something upsets me I try to keep my emotions in balance.
_____ 10. When I feel inadequate in some way, I try to remind myself that feelings of inadequacy are shared by most people.
_____ 11. I'm intolerant and impatient towards those aspects of my personality I don't like.
_____ 12. When I'm going through a very hard time, I give myself the caring and tenderness I need.
_____ 13. When I'm feeling down, I tend to feel like most other people are probably happier than I am.
_____ 14. When something painful happens I try to take a balanced view of the situation.
_____ 15. I try to see my failings as part of the human condition.
_____ 16. When I see aspects of myself that I don't like, I get down on myself.
_____ 17. When I fail at something important to me I try to keep things in perspective.

______ 18. When I'm really struggling, I tend to feel like other people must be having an easier time of it.

______ 19. I'm kind to myself when I'm experiencing suffering.

______ 20. When something upsets me I get carried away with my feelings.

______ 21. I can be a bit cold-hearted towards myself when I'm experiencing suffering.

______ 22. When I'm feeling down I try to approach my feelings with curiosity and openness.

______ 23. I'm tolerant of my own flaws and inadequacies.

______ 24. When something painful happens I tend to blow the incident out of proportion.

______ 25. When I fail at something that's important to me, I tend to feel alone in my failure.

______ 26. I try to be understanding and patient towards those aspects of my personality I don't like.

GAD 7 Questionnaire

Over the last 2 weeks, how often have you been bothered by the following problems? *(Use "✔" to indicate your answer"*	Not at all	Several days	More than half the days	Nearly every day
1. Feeling nervous, anxious or on edge	0	1	2	3
2. Not being able to stop or control worrying	0	1	2	3
3. Worrying too much about different things	0	1	2	3
4. Trouble relaxing	0	1	2	3
5. Being so restless that it is hard to sit still	0	1	2	3
6. Becoming easily annoyed or irritable	0	1	2	3
7. Feeling afraid as if something awful might happen	0	1	2	3

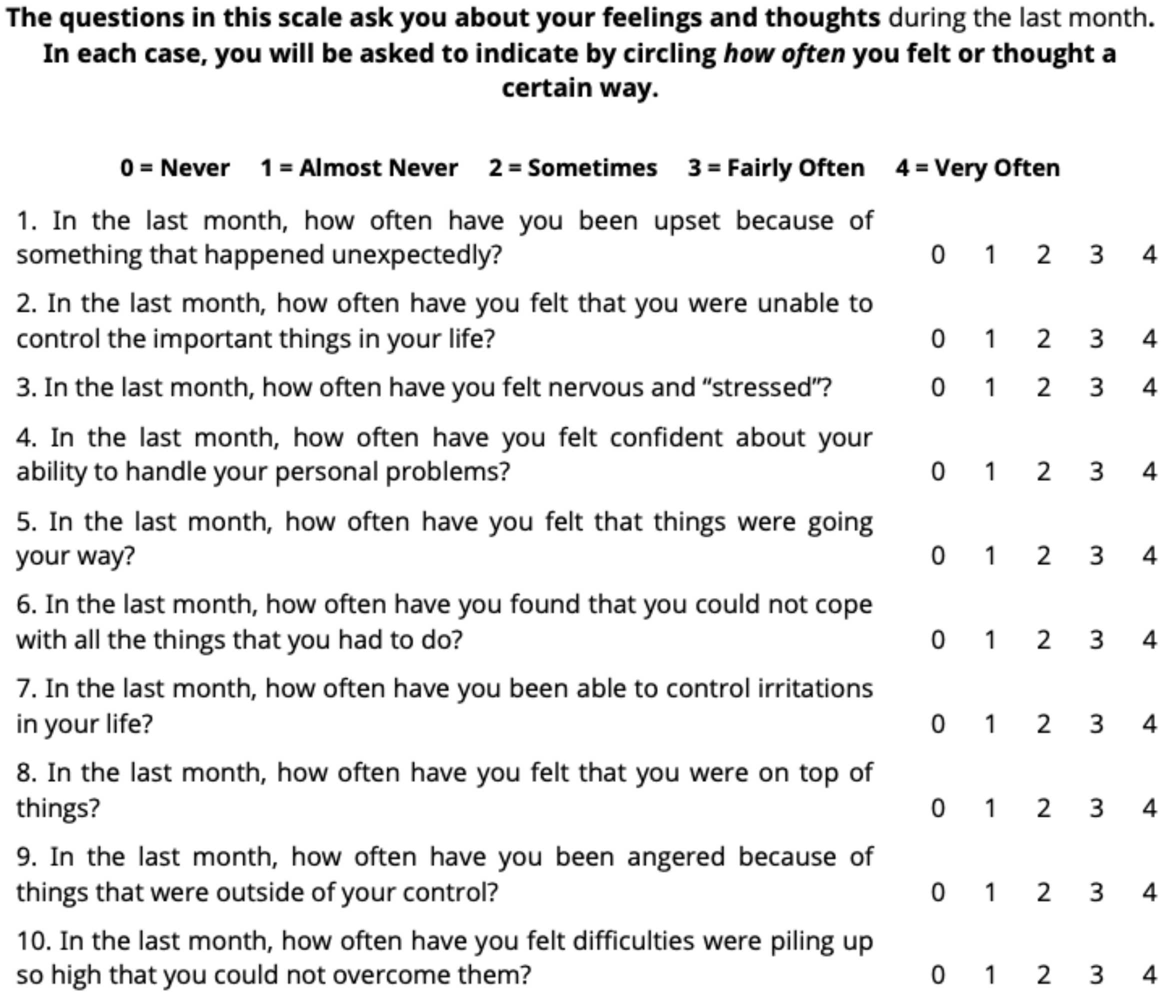
If you checked off any problems, how difficult have these problems made it for you to do your work, take care of things at home, or get along with other people?

Not difficult at all	**Somewhat difficult**	**Very difficult**	**Extremely difficult**
☐	☐	☐	☐

PHQ9 Questionnaire

Over the last 2 weeks, how often have you been bothered by any of the following problems? *(Use "✔" to indicate your answer")*	**Not at all**	**Several days**	**More than half the days**	**Nearly every day**
1. Little interest or pleasure in doing things	0	1	2	3
2. Feeling down, depressed, or hopeless	0	1	2	3
3. Trouble falling or staying asleep, or sleeping too much	0	1	2	3
4. Feeling tired or having little energy	0	1	2	3
5. Poor appetite or overeating	0	1	2	3
6. Feeling bad about yourself — or that you are a failure or have let yourself or your family down	0	1	2	3
7. Trouble concentrating on things, such as reading the newspaper or watching television	0	1	2	3
8. Moving or speaking so slowly that other people could have noticed? Or the opposite — being so fidgety or restless that you have been moving .around a lot more than usual	0	1	2	3
9. Thoughts that you would be better off dead or of hurting yourself in some way	0	1	2	3

Perceived Stress Scale



The questions in this scale ask you about your feelings and thoughts during the last month**. In each case, you will be asked to indicate by circling *how often* you felt or thought a certain way.**

0 = Never 1 = Almost Never 2 = Sometimes 3 = Fairly Often 4 = Very Often

1. In the last month, how often have you been upset because of something that happened unexpectedly?	0	1	2	3	4
2. In the last month, how often have you felt that you were unable to control the important things in your life?	0	1	2	3	4
3. In the last month, how often have you felt nervous and "stressed"?	0	1	2	3	4
4. In the last month, how often have you felt confident about your ability to handle your personal problems?	0	1	2	3	4
5. In the last month, how often have you felt that things were going your way?	0	1	2	3	4
6. In the last month, how often have you found that you could not cope with all the things that you had to do?	0	1	2	3	4
7. In the last month, how often have you been able to control irritations in your life?	0	1	2	3	4
8. In the last month, how often have you felt that you were on top of things?	0	1	2	3	4
9. In the last month, how often have you been angered because of things that were outside of your control?	0	1	2	3	4
10. In the last month, how often have you felt difficulties were piling up so high that you could not overcome them?	0	1	2	3	4

Quality of Life Scale

Please read each item and circle the number that best describes how satisfied you are at this time. Please answer each item even if you do not currently participate in an activity or have a relationship. You can be satisfied or dissatisfied with not doing the activity or having the relationship.

	Delighted	Pleased	Mostly Satisfied	Mixed	Mostly Dissatisfied	Unhappy	Terrible
1. Material comforts home, food, conveniences, financial security	7	6	5	4	3	2	1
2. Health - being physically fit and vigorous . . .	7	6	5	4	3	2	1
3. Relationships with parents, siblings & other relatives- communicating, visiting, helping . . .	7	6	5	4	3	2	1
4. Having and rearing children	7	6	5	4	3	2	1
5. Close relationships with spouse or significant other	7	6	5	4	3	2	1
6. Close friends	7	6	5	4	3	2	1
7. Helping and encouraging others, volunteering, giving advice	7	6	5	4	3	2	1
8. Participating in organizations and public affairs	7	6	5	4	3	2	1
9. Learning- attending school, improving understanding, getting additional knowledge . .	7	6	5	4	3	2	1
10. Understanding yourself - knowing your assets and limitations - knowing what life is about . .	7	6	5	4	3	2	1
11. Work - job or in home	7	6	5	4	3	2	1
12. Expressing yourself creatively	7	6	5	4	3	2	1
13. Socializing - meeting other people, doing things, parties, etc	7	6	5	4	3	2	1
14. Reading, listening to music, or observing entertainment	7	6	5	4	3	2	1
15. Participating in active recreation	7	6	5	4	3	2	1
16. Independence, doing for yourself	7	6	5	4	3	2	1

Appendix F: Prevalence of Chronic Conditions

Table 11

Prevalence of Chronic Conditions in the Children of Caregivers in the Sample

Chronic condition	**N (%)**
Kidney/bladder disorders	41 (20%)
Digestive system disorders	39 (19%)
Anxiety	29 (14.1%)
Autism Spectrum disorders	28 (13.7%)
Pain disorders	20 (9.8%)
Epilepsy/Seizures	20 (9.8%)
Arthritis	20 (9.8%)
Depression	19 (9.3%)
Attention Deficit disorders	17 (8.3%)
Hypermobility/Ehrler’s Danlos Syndrome	17 (8.3%)
Diabetes	14 (6.8%)
Genetic disorders	11 (5.4%)
Developmental delay/Intellectual disability	11 (5.4%)
Cerebral palsy	10 (4.9%)
Asthma	8 (3.9%)
Eating disorders	6 (2.9%)
Bipolar disorder	6 (2.9%)
Chronic fatigue syndromes	6 (2.9%)
Heart disorders	6 (2.9%)
PTSD	6 (2.9%)
Spinal disorders	5 (2.4%)
Coeliac disease	4 (2%)
Oppositional defiant disorder	4 (2%)
Autoimmune/inflammatory disorders	4 (2%)
Postural orthostatic tachycardia syndrome	4 (2%)
Dyspraxia	4 (2%)
Other disorders	45 (22.2%)

Note: The category ‘other disorders’ encompasses:
(*N* = 3): Borderline Personality Disorder, Skin disorders, Hydrocephalus, Psychosis, Hearing or sight loss, Sleep disorders, Sensory processing disorder
(*N* = 2): Attachment disorders, Allergies, Trauma, Fibromyalgia, Brain injury/Functional neurological disorder
(*N* = 1): Motor tic disorder, lung defect, Tourette’s syndrome, Emotional dysregulation, Hypothyroidism, Obsessive compulsive disorder, Stroke, Nervous system dysregulation, Familial cold autoinflammatory syndrome, Precocious puberty, immune thrombocytopenic purpura, Pituitary tumour, PICA, Paraplegia.

Appendix G. Summary of Author Guidelines: Behavioural Research and Therapy

Article structure

Subdivision - unnumbered sections

Divide your article into clearly defined sections. Each subsection is given a brief heading. Each heading should appear on its own separate line. Subsections should be used as much as possible when cross-referencing text: refer to the subsection by heading as opposed to simply 'the text'.

Appendices

If there is more than one appendix, they should be identified as A, B, etc. Formulae and equations in appendices should be given separate numbering: Eq. (A.1), Eq. (A.2), etc.; in a subsequent appendix, Eq. (B.1) and so on. Similarly for tables and figures: Table A.1; Fig. A.1, etc.

Essential title page information

- Title. Concise and informative: Titles are often used in information-retrieval systems. Avoid abbreviations and formulae where possible.
- Author names and affiliations: Please clearly indicate the given name(s) and family name(s) of each author and check that all names are accurately spelled. You can add your name between parentheses in your own script behind the English transliteration. Present the authors' affiliation addresses (where the actual work was done) below the names. Indicate all affiliations with a lowercase superscript letter immediately after the author's name and in front of the appropriate address. Provide the full postal address of each affiliation, including the country name and, if available, the e-mail address of each author
- Corresponding author: Clearly indicate who will handle correspondence at all stages of refereeing and publication, also post-publication. This responsibility includes answering any future queries about Methodology and Materials. Ensure that the e-mail address is given and that contact details are kept up to date by the corresponding author.
- Present/permanent address: If an author has moved since the work described in the article was done, or was visiting at the time, a 'Present address' (or 'Permanent address') may be indicated as a footnote to that author's name. The address at which the author actually did

the work must be retained as the main, affiliation address. Superscript Arabic numerals are used for such footnotes.

Abstract

A concise and factual abstract is required with a maximum length of 200 words. The abstract should state briefly the purpose of the research, the principal results and major conclusions. An abstract is often presented separately from the article, so it must be able to stand alone. For this reason, References should be avoided, but if essential, then cite the author(s) and year(s). Also, non-standard or uncommon abbreviations should be avoided, but if essential they must be defined at their first mention in the abstract itself.

Keywords

Immediately after the abstract, provide a maximum of 6 keywords, to be chosen from the APA list of index descriptors. These keywords will be used for indexing purposes.

Abbreviations

Define abbreviations that are not standard in this field in a footnote to be placed on the first page of the article. Such abbreviations that are unavoidable in the abstract must be defined at their first mention there, as well as in the footnote. Ensure consistency of abbreviations throughout the article.

Tables

Please submit tables as editable text and not as images. Tables can be placed either next to the relevant text in the article, or on separate page(s) at the end. Number tables consecutively in accordance with their appearance in the text and place any table notes below the table body. Be sparing in the use of tables and ensure that the data presented in them do not duplicate results described elsewhere in the article. Please avoid using vertical rules and shading in table cells.

References

Citation in text

Please ensure that every reference cited in the text is also present in the reference list (and vice versa). Any references cited in the abstract must be given in full. Unpublished results and personal communications are not recommended in the reference list, but may be mentioned in the text. If these references are included in the reference list they should follow the standard reference style of the journal and should include a substitution of the publication date with either 'Unpublished results' or 'Personal communication'. Citation of a reference as 'in press' implies that the item has been accepted for publication.

Web references

As a minimum, the full URL should be given and the date when the reference was last accessed. Any further information, if known (DOI, author names, dates, reference to a source publication, etc.), should also be given. Web references can be listed separately (e.g., after the reference list) under a different heading if desired or can be included in the reference list.

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