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Review Article

Title

Self-perceived Burden for People with Life-threatening Illness: A Qualitative Systematic Review

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Abstract

Context: The perception of being a burden to others is a significant concern for people with life-threatening illness. It is unclear what underpins the concept of “self-perceived burden”.

Objective: To appraise and integrate primary evidence underpinning the concept of self-perceived burden with respect to their informal caregivers among adult patients with life-threatening illness.

Methods: This is a systematic review and a thematic synthesis of qualitative primary data. MEDLINE, Embase, PsycINFO, CINAHL, and Web of Science were searched in September 2021, supplemented by hand searching of textbooks and web search engines, peer-review journals, and contact with experts. Qualitative studies were included if they provided primary data of adult patients’ (with life-limiting illness) self-perceived burden toward their informal caregivers. Studies were appraised using the Critical Appraisal Skills Programme checklist.

Results: Nine studies were included, reporting on 219 patients who mostly had advanced life-threatening illness and needed physical assistance for daily activities. Self-perceived burden is a highly subjective perception contrasting self and/or informal caregivers in the past, present, and future. Patients develop self-perceived burden feeling uncertain about caregivers’ perceptions and increasing future burden. Self-perceived burden interacts with other factors of surrounding complex balances (such as care needs, reality, and identity) which would change over time, and patients’ reactions to self-perceived burden at the sacrifice of their wishes may conversely increase the total suffering.

Conclusion: Self-perceived burden is not a static perception but a fluctuating and complex 'process' based on uncertainty. More diverse understandings and following interventions to achieve a better balance of care should be sought.

Key words

Burden, palliative care, qualitative, review

Key Messages

Self-perceived burden is a highly subjective perception based on uncertainty towards caregivers and the future. The perception is not either static or temporary but a fluctuating 'process' in complex balances among numerous factors, therefore it should be continuously explored in view of the theory of complexity.

Running Title

Patients' self-perceived burden to others

Background

The concern of 'being a burden to others' is common among people with life-threatening illnesses.^{1 2} It has been largely explored more from the perspective of caregivers' rather than patients',^{3 4 5} although caregivers may not accurately understand patients' feelings.⁶

Cousineau et al. developed the concern of 'being a burden to others' into the concept of 'self-perceived burden (SPB)'. It is defined as *'a multidimensional construct arising from the care recipient's feelings of dependence and the resulting frustration and worry, which then may lead to negative feelings of guilt at being responsible for the caregiver's hardship'*.⁷ This was later revised by McPherson et al., reflecting that dependency is not inevitable, and highlighting the diminishing sense of self: *'empathic concern engendered from the impact on others of one's illness and care needs, resulting in guilt, distress, feelings of responsibility, and diminished senses of self'*.⁸

Cousineau's Self-Perceived Burden Scale⁷ has demonstrated the high prevalence of SPB among advanced cancer, post-stroke, and amyotrophic lateral sclerosis (ALS) patients, reported as 28-73%,^{9 10 11} 66-70%,^{12 13 14} and 67%¹⁵ respectively. Demographic factors including female sex, younger age, and poor financial status are associated with higher SPB.¹⁶ The prevalence of SPB increases among cancer patients as death approaches¹⁷ but decreases among patients after stroke.¹⁸

A prior systematic review identified SPB as a key factor in many aspects of palliative care, including advance care planning, place of death, and quality of life.¹⁹ However, the review found only one qualitative study primarily exploring SPB. A further review found that SPB drives patients' wish to hasten death, although the study focused on SPB in the context of suicidal thoughts.²⁰

Investigation into SPB lacks an understanding of the theoretical concepts that underpin it. It is needed to integrate what qualitative data have informed about SPB to develop further research and clinical implementations to support patients and their informal caregivers. Therefore, this review aimed to appraise and integrate primary evidence underpinning the concept of SPB among adult patients with life-threatening illness with respect to their informal caregivers. The review question is 'What makes people with life-threatening illness develop SPB regarding their informal caregivers and how do they experience the perception?'

Method

Design

This review was reported according to the Enhancing transparency in reporting the synthesis of qualitative research (ENTREQ) statement.²¹ Data was synthesised using thematic synthesis, a widely used method of synthesising qualitative studies.²² Methods to synthesise qualitative studies have been increasingly developed and have gained importance to deal with expanding needs of building better understandings than just the sum of individual findings.²³ A preliminary search suggested that there might be few studies with a primary aim of studying SPB. Thematic synthesis was therefore chosen, since its interpretative process enables 'go beyond' the findings of the studies with heterogeneous aims and develop relevant findings to the specific research aim of this review.²² A.S. conducted the literature searching, data extraction, quality assessment, and analysis. The second reviewer (A.O.) played a role as a second checker and had discussions until agreement was achieved when there were any disagreements or

uncertainty. The third reviewer (R.H.) provided oversight and consultation when necessary.

Search strategy

MEDLINE, Embase, PsycInfo, CINAHL, and Web of Science were searched in September 2021 (See terms in Supplemental Appendix 1). Librarians were consulted and provided double-check when the search strategies were developed. We supplemented this with hand searching of textbooks, citation searching using web search engines, and hand searching of relevant journals. Since SPB might be too ambiguous to be sufficiently found through these searching methods, experts who had published qualitative studies on SPB were asked whether they knew any studies that met the criteria. The eligibility criteria were developed along with Sample, Phenomenon of Interest, Design, Evaluation, and Research type.²⁴ (Table 1) The eleven most common life-threatening illnesses were selected from the estimation of global serious health-related suffering.²⁵ Studies were included if participants were aged 18 or older and if more than 50% of the participants reported a diagnosis of one of the life-threatening illnesses (Details of the illnesses are presented in Supplemental Appendix 2). Caregivers were defined as “*unpaid, informal providers of one or more physical, social, practical, and emotional tasks. In terms of their relationship to the patient, they may be a friend, partner, ex-partner, sibling, parent, child, or other blood or non-blood relative*”.²⁶ Texts in line with four components of SPB defined by McPherson et al.⁸ (Figure 1) were regarded as data of SPB.

Quality assessment

Data were extracted from the results/findings sections of papers into NVivo (Released 2020). Key factors including aims, setting, design, participants, and major findings were

tabulated using a Microsoft Excel spreadsheet to compare heterogeneous studies. Quality appraisal was conducted using the Critical Appraisal Skills Programme (CASP) checklist to deeper understand the overall study methodology and findings from the general factors of qualitative study appraisal such as methodological appropriateness of data collection, analysis, reporting, and relevance to this review.²⁷ In addition to the three categories 'yes', 'no' or 'can't tell', a response 'somewhat' was added.²⁸

Analysis

Data were synthesised using NVivo and three stages of thematic synthesis were conducted.²²

Step I. Coding texts "line-by-line": Included papers were read and re-read being taken notes, highlighted, and coded. Texts that were not associated with the research questions were not extracted (for example, texts mentioning relationships solely between patients and health care professionals).

Step II. Developing descriptive themes: Codes were grouped, connected with lines, and reallocated. Even if the codes could be applied to several clusters, they were allocated to best fit the aim and objectives of this review. The figure did not become a simple hierarchical tree because the structure of SPB was found to be complex. Fourteen clusters of codes were identified and given descriptive themes that illustrate the characteristics of the clusters.

Step III. Generating analytical themes: Descriptive themes were compared and explored. The analytical themes were developed inductively and iteratively until the analytical themes went beyond the contents of the original texts and descriptive themes but sufficiently illustrated the overarching concept.

Results

Included studies

Database searching identified 3301 papers, and 2291 papers remained following deduplication (see PRISMA flowchart Figure 2). Following title and abstract screening, 215 papers remained, with an additional 26 papers identified through other searching methods. After full-text papers were retrieved and eligibility assessed, nine papers that met the inclusion criteria were retained for analysis.

The nine studies from eight substantive research studies reported a total 219 patients. Studies were conducted in Australia (n=3), the Republic of Ireland (n=2), Switzerland (n=1), Japan (n=1), Canada (n=1), and the UK (n=1). Four studies focused on advanced cancer, three on ALS, one on chronic renal failure, and one on multiple conditions with over half of the sample having a cancer diagnosis. The majority of the participants had advanced life-threatening illness and needed physical assistance for daily activities. Two of the included studies involved exploring SPB in their research aims. Study summaries are presented in Table 2.

Study quality

The quality of the studies greatly varied, with five scoring 'yes' for at least half of the items. Inadequate reporting lowered study quality score more than methodological issues (see Table 3).

Data synthesis & main findings

The analysis developed 14 clusters containing 51 codes, from which six analytical themes were generated (See Supplemental Appendix 3): 1) *Sense of burden on informal caregivers and patients*, 2) *SPB is experienced within lifelong relationships between patients and informal caregivers*, 3) *SPB as a result of comparing hardships and lost*

opportunities, 4) SPB is based on uncertainty, 5) Fluctuating SPB, and 6) Reactions to SPB are part of the process. Illustrative quotes are provided with brief demographic data.

1) *Sense of burden on informal caregivers and patients.* The data revealed burdens on informal caregivers and on patients, and patients felt physical, psychological, and social burdens towards their informal caregivers.

Physical burdens generally consisted of housework (e.g. laundry, cleaning, or shopping)⁸³⁰³¹³⁶ and care for the patient (e.g. dressing clothes, managing medicines or medical devices, or taking the patient to hospital).⁸³⁰³⁴ The patients were more susceptible to being concerned about physical burdens if the informal caregivers were old,³⁶ ill,⁸³¹³⁵ or already exhausted.³⁴

Psychological burdens (some of which were also categorised as spiritual burdens) were associated with worrying about patients all the time,⁸²⁹ having “difficult conversations” (such as about how their condition would deteriorate),⁸²⁹³⁴ and with the dying process (e.g. accepting the patients’ illness⁸ or witnessing uncontrolled symptoms).⁸³¹ As a result, a variety of informal caregivers’ emotions such as sadness,²⁹ anxiety,⁸ or worry²⁹³² were provoked, that were felt by the patient to be burdensome.³³

Social burdens included care demands that impinged on informal caregivers’ daily lives and other commitments such as housework, childcare, or paid work,⁸²⁹³⁰³¹³²³⁵³⁶ especially when the caregivers were younger than the participants and experienced impact on their lives.³²³⁶

The participants were also worried about increasing future burdens⁸²⁹³¹³⁴ even after the participant’s death.⁸²⁹³⁴³⁵

These perceived burdens on caregivers caused significant distress for patients.²⁹³³ The patients consequently described feelings of responsibility,⁸ guilt,⁸³⁴³⁶ anger,⁸³³ or fear.³³

³⁴ They also felt dependent on caregivers describing themselves as useless,^{8,34} failure,³⁴ or having loss of control,^{8,31,34} and the feelings sometimes made patients feel isolated or hopeless.³⁴ Patients also described how hard it was to witness informal caregivers' struggling with burdens,^{8,34} and this gave a corresponding sense of burden to patients.

'It upsets me to see her [wife] upset' [male, ALS]²⁹

2) *SPB is experienced within lifelong relationships between patients and informal caregivers.* Family contexts influenced how the patients perceived the situation. Role shifts from caregiver and supporter in life to become patient and recipient of informal care framed SPB, particularly for older people and parents of young children. Older participants had a sense of accomplishment having raised children and grandchildren, while younger patients (especially those with young children) expressed strong concerns of imposing early hardships on informal caregivers.^{8,32,34} The role as a parent of young children was described as an unfulfilled task, and the perceived incompleteness as a burden on others.⁸ Similarly, traditional gender roles were also described as a burdensome role shift.^{8,34}

The pre-existing positive relationship between patient participants and their informal caregivers had built a sense of appreciation, particularly for enabling the patient to be cared for at home.^{29,32,33,34,36} This appreciation amplified patient concern for the caregiver,^{29,31} and consequently patients prioritised their caregivers over their own concerns and wellbeing.

'So that's my major concern, is not for myself, but the extra burden [returning home is] placing on [his wife]'. [male]³⁵

3) *SPB as a result of comparing hardships and lost opportunities.* The participants also acknowledged that they have (or expect to) become physically and cognitively impaired,

with reducing independence^{8 32 33 36} or dignity.³¹ The altered self fundamentally challenged identity.^{31 34}

'My whole life, I only worked and always took care [of others] myself. ... Then after this it's just difficult, if you always have to have other people. Having to be a burden. ... That I've never liked.' [elderly, female, cancer]³⁴

Patients reported caregiver burden as the difference between the care tasks they had to perform and what they might otherwise have done with their time, as like the theory of the opportunity cost. Additional burden was described from in the caregiving demands being greater than the caregiver capacity.³¹

'All the pain and anguish and the anger and everything that I went through, and I'd hate to inflict that on them [family]. It would be unfair to them really.' [female, ALS]³³

4) *SPB is based on uncertainty.* First, uncertainty regarding the future was consistently reported. The illnesses had already brought significant changes in most of the participants' lives, and they were aware that transitions towards the terminal stage would bring further significant deterioration.³⁶ Some participants were eager to know the likely future illness trajectory so that they could prepare for the sake of informal caregivers.³³ Lack of such knowledge provoked fear and anxiety among both patients and informal caregivers.^{29 32 33}

Second, patients were not sure whether informal caregivers were coping with the care demands upon them and how they would react to the further change.^{29 31 32} The uncertainty concerning informal caregivers was greater when they did not express their feelings to patients.³¹

'But watching them, watch you, decline and die, I can't imagine what that's like. Because I'm not them, I'm me, I can't imagine how they feel and it's not, it's not pleasant.' [61-70 years old, female, hospice in-patient]³¹

Patients felt SPB even without any sense of blame for having developed a serious illness, despite having informal caregivers who did not express negative reactions to their role, and although they felt well supported.⁸ Therefore, SPB is a subjective perception that is not always based on expressed burden from caregivers or deficits in informal care.

5) *Fluctuating SPB and complex balances.* Participants were concerned to ask for care without excessively troubling others, although it was difficult to find a good "equilibrium" between asking appropriately and fearing to cause burden as the situation changed. For example, when patients tried to consider better balance in decision making around place of care, SPB interplayed with what patients regarded as 'needed', 'feasible', and 'right' in view of relational impacts with informal caregivers.³⁵ Further, adjustment to new roles was crucial, although patients and informal caregivers continuously needed to become used to the deteriorating situations. Lack of choice in their care led to acceptance, resignation, and even a sense of being at a loss.^{8 32}

'Well she's got a few health issues and I don't want to put too much on her, you know? If it's, if everything was manageable, yeah I'd go home, yeah. But if it's not practical I'll have to stay here.' [male, 61-70 years old, pancreatic cancer]³⁵

Death-hastening thoughts were also partly driven by the nature of SPB: some participants associated suicidal thoughts with lifting burden from informal caregivers, but they were also conscious that death-hastening actions may cause more burdens for informal caregivers.^{29 33 34}

6) *Reactions to SPB are part of the process.* Patients took various actions to manage SPB and the reactions were also embedded in the process of SPB. Some measures apparently worked well, while others may increase burdens or other unintended negative effects.

Firstly, patients reported 'trying to interact with informal caregivers and burdens', including finding positive meanings in burdens or entitlement for the care,^{8 36} discussing and making decisions together,^{8 34 36} making the most of what they currently can do,^{8 29 31 34 36} or preparing for the future burdens.^{8 29} These actions work well when patients and informal caregivers worked together on finding a better balance for them both. Further, care recipients were also care providers for both themselves and informal caregivers,^{31 32 33} and they tried to 'protect' informal caregivers from burdens.³⁵

'I'm glad that the family explained the situation to me, telling me that my condition was getting no better, that the family was struggling to care for me on their own, and that it would be unfeasible for me to go on living at home. I am glad that we could discuss my care situation together with my family.' [cancer]³⁶

The second approach was 'placing a distance from informal caregivers and burdens: they conversely kept away from informal caregivers or burdens by suppressing their own feelings,^{8 29 34 36} trying not to think about the burden,^{8 29 32 34 36} hoping for sudden death,^{8 31 32 33 34} reducing contacts,^{8 29 31 34 36} or maintaining normality.^{8 29} These strategies generally did not ease SPB, and may bring other hardships because patients sacrificed their own wishes or care needs. Of some concern, these strategies can bring a sense of separation between patients and informal caregivers. Patients also tried to conceal changes in their wellbeing to maintain normality and prevent informal caregivers' distress.^{8 29}

The third approach was passive inaction to reduce SPB. This included not trying new care or treatment plans to avoid extra burdens on informal caregivers; withholding potentially life-sustaining treatments hoping not to add anything 'unnecessary' or not to just 'drag on' their lives.³³ Other participants 'accepted' to be taken care of with a sense of resignation.

'Either you accept it or you're in bad humor. Mind you, I was in a bad humor for a while because it was getting on my nerves. Eventually it got better. But again there is nothing you can do about it. I try to accept it. I still feel a bit, I always will be a burden, but I got used to it, I have to accept it.' [cancer]⁸

Discussion

The six themes presented above are in line with the McPherson definition of SPB:⁸ patients feel that illness-altered selves imposed (or will impose) burdens on their informal caregivers, leading them to experience negative feelings such as of frustration, guilt, responsible, and also feel themselves burdened. Patients feel SPB when comparing themselves with their informal caregivers and also their previous/future self, and they describe violation of their capability and identity. The current review also revealed inevitable uncertainty and complex balances illustrating SPB as a fluctuating process. Uncertainty of SPB was perceived in light of caregivers and the future: the patient's sense of burden may not be borne out by the reality or perception of the caregiver, and uncertainty surrounding anticipating future deterioration and burden also fuel patients' current SPB. The finding is compatible with that a prior study identified uncertainty as a dominant theme in end-of-life patients' perception of care and illness experiences,³⁷ and the fundamental uncertainty indicates that SPB might be hard for patients even to face

with and clearly capture. SPB is also not either a static or temporary phenomenon but a process which would change over time. Despite patients' actions to alleviate the sense of being a burden, distress may not sufficiently be alleviated because the sense itself may fluctuate and the reactions may bring other hardships such as separation from caregivers. Indeed, the subjective perception of SPB should be continuously addressed as it informs patient decision-making.

SPB may be understood within a 'complex adaptive system' (in which a group of individuals act without clear predictability).³⁸ Prior research into complexity in palliative care has revealed common factors with this review, including family caregiving, prior experience, and adaptive processes through uncertainty.^{39 40 41} The theory of complexity would support a better balance of care for both patients and their informal caregivers.

There are a number of limitations to our review. Firstly, a possibility of bias underlies since this review was primarily conducted by one author. However, the other authors were consulted when necessary at any points of the review process and discussion was done until the agreement was achieved, supplemented by contacts with librarians and external experts. The whole results were agreed by all of the authors to improve the trustworthiness. Second, the cultural and clinical/research background of the authors might have narrowed interpretation, although discussions and transparency of the methods were addressed to improve reflexivity. Reliance on the pre-existing definition of SPB may have restricted findings as well, but this review would trigger conceptual understandings of SPB. Third, papers may have been excluded due to our limit of papers in English.

We strongly recommend further research to understand the concept of SPB in non-Western countries, and drawing from populations with more diverse diagnoses of life-

threatening illness. Beyond conceptualisation and measurement, interventions to identify and reduce SPB are needed. Considering the subjectivity, uncertainty, and changeability, interventions that improve patients' cognitive coping skills might work well.

Conclusion

This review developed the theoretical concept of SPB among adult patients with life-threatening illness revealing surrounding uncertainty and complex balances. SPB was also presented to be a process which may change over time as the patients' conditions would inevitably deteriorate, therefore health care professionals and researchers should keep addressing SPB in line with individual patient's illness experiences. We propose the theory of complexity would support to ease patients' whole suffering including SPB, while measures that only deal with SPB might increase other hardships. Qualitative aspects of SPB from more diverse cultures and illness trajectories need to be explored to gain more comprehensive understandings. Since SPB is a major concern for people with life-threatening illness and an important factor of numerous relevant factors in palliative care, it should be continuously addressed to achieve better balance of care and quality of life of patients with life-threatening illness and their informal caregivers.

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All relevant data are presented in the manuscript and supplemental appendices. Other data are available if requested to the corresponding author.

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Table 1. Summary of eligibility criteria

	Inclusion	Exclusion
Sample	<ul style="list-style-type: none"> • Adult patients aged 18 or over • More than 50 % of the patients (or survivors) who participated in the study were identified to have any of the following life-threatening illnesses <ul style="list-style-type: none"> ○ malignant neoplasms ○ cerebrovascular disease ○ lung disease ○ dementia ○ liver disease ○ heart disease ○ tuberculosis ○ human immunodeficiency virus disease ○ renal failure ○ atherosclerosis ○ degenerative central nervous system disease 	<ul style="list-style-type: none"> • Caregivers' data as patient proxies
Phenomenon of interest	<ul style="list-style-type: none"> • SPB as a result of the illnesses identified above • Investigating the relationship between patients and informal caregivers • Data reported in line with four components of SPB. (Figure 1) 	<ul style="list-style-type: none"> • Investigating the relationship only between patients and people other than informal caregivers (such as health care professionals)

<p>Design, evaluation, and research type</p>	<ul style="list-style-type: none">• Original qualitative or mixed methods studies which include qualitative methods in the text• Including relevant rich descriptions to identify key concepts which underpin the idea of SPB• Published in peer-reviewed journals in 2001 or later• Written in English	<ul style="list-style-type: none">• Not using qualitative methods for data collection or data analysis• Case reports of a patient, reviews, editorials, letters, or conference abstracts
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Table 2. Summary of the included studies

Study	Aims	Country	Setting	Design		
				Sampling method	Data collection method	Methodological approach
Ando, H <i>et al.</i> (2019) ²⁹	To explore how the WHOQoL aspect of 'concerns' is relevant in the lives of people with ALS.	UK	in community	purposive sampling	in-depth semi-structured interview	TA
Ashby, M <i>et al.</i> (2005) ³⁰	To understand why patients chose to stop or start dialysis.	Australia	4 at nephrology units in-patients of tertiary hospitals and 7 in community	referral-based sampling	semi-structured interview	GT
Broom, A <i>et al.</i> (2012) ³¹	To explore perspectives and experiences in the last few weeks of life including those related to families.	Australia	at hospice	not clearly stated (possibly purposive sampling)	in-depth relatively unstructured interview	reference not mentioned
Foley, G <i>et al.</i> (2016) ³²	To explore how ALS patients experience and make decisions regarding care.	Republic of Ireland	32 at home, 1 at nursing home, and 1 at hospice	theoretical sampling	in-depth unstructured interview	GT

Foley, G <i>et al.</i> (2014) ³³	To illuminate how life courses shape responses of people with ALS to impending mortality and decision-making about their care.	Republic of Ireland	32 at home, 1 at nursing home, and 1 at hospice	theoretical sampling	in-depth unstructured interview	GT
Gudat, H <i>et al.</i> (2019) ³⁴	To understand what terminally ill patients think and intend when experiencing a wish to die.	Switzerland and	at PC hospital, PC ward of an oncology department, and in community	theoretical sampling but sometimes convenience	semi-structured interview	combined GT and IPA
MacArthey, J. I. <i>et al.</i> (2016) ³⁵	To address how participants' experiences of home care and in-patient palliative hospital care affected where they might want to die.	Australia	at specialist PC unit	purposive sampling	semi-structured interview	dialogical interviewing approach and sociological interpretive methodology
McPherson, C. J. <i>et al.</i> (2005) ⁸	To explore SPB from the perspective of terminally ill patients.	Canada	14 at home and 1 at long-term care facility	purposive sampling	interview with less-structured 3 major lines of questioning	IPA

Oeki, M <i>et al.</i> (2020) ³⁶	To elucidate advanced cancer patients' strategies to cope with their SPB.	Japan	at PC unit	purposive sampling	semi-structured interview	CA
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Table 2. Summary of the included studies (continued)

Study	Participants				Study participants other than patients	Details of primary caregivers	Sections extracted for analysis	Major findings
	N (male/female)	Age	Clinical characteristics	ADL				
Ando, H <i>et al.</i> (2019) ²⁹	26 (14/12)	32-84 y/o (mean: 64)	ALS, mean illness duration 20 months	ALSFR-S: median 38, inter-quartile range 13	none, but 6 participants attended the interview with a companion	not stated	All	Significance of significant others led to more stress on significant others' current and future well-being, and the participants tried to decrease the impacts caused by the disease even putting themselves aside.
Ashby, M <i>et al.</i> (2005) ³⁰	11 (not stated)	57-89 y/o (median: 77)	CRF, 4 discontinued dialysis and 7 abated dialysis	not stated	5 participants' spouses	not stated	'Desire not to be a burden'	'Not want to be a burden to their families' was an overarching theme regarding the decision to terminate or refuse dialysis. One of the significant factors that affected the feeling was ageing.

Broom, <i>A et al.</i> (2012) ³¹	20 (11/9)	41-80 or over y/o (a participa nt did not reveal age)	hospice in- patients, mostly advanced cancer	not stated	none	11 currently married, 3 divorced, 2 widowed, 1 de facto partnership, 1 never married, 2 concealed their status. 19 had children.	The strain on families/	The guilty feelings of being a burden and a strain to their families at home urged them to move to the hospice, which they found a relief and a solution for their families.
Foley, G34 <i>et al.</i> (2016) ³²	34 (17/17)	37-81 y/o (median: 60-69)	ALS, mean duration 31 months	32 needed assistance for ADL, 29 used assistive devices, 8 had gastrostomy feeding and/or NIV	none, significant others were present at the 9 interviews	22 spouses/partners, 4 children, 4 sisters, 1 nephew, 1 daughter-in-law, 2 lived at care facilities were not mentioned	All	The participants appreciated the care from their families but also greatly suffered from the feeling of imposing a burden on their families, which made the participants feel obliged to their families.
Foley, G34 <i>et al.</i> (2014) ³³	34 (17/17)	37-81 y/o (median: 60-69)	ALS, mean duration 31 months	32 needed assistance for ADL, 29 used assistive devices, 8 had gastrostomy	none, the significant others were present at the 9 interviews	22 spouses/partners, 4 children, 4 sisters, 1 nephew, 1 daughter-in-law, 2 lived at care	'Family: Context to Decision Making'	Family was a significant context of decision making, and they struggled to find a good balance between 'being a burden' to their family and 'resigning to be dependent on them'.

				feeding and/or NIV		facilities were not mentioned		
Gudat, H <i>et al.</i> (2019) ³⁴	62 (not stated)	not stated	11 advanced cancer, 11 organ failure, 10 degenerative neurological disease, 11 frailty	not stated	family caregivers (one of the patient's close relatives) and professionals	not stated	All (but the texts on ICs was excluded)	The participants wished to hasten death to unburden others but also decide not to hasten death to unburden others, which made them stuck and suffered between those feelings.
MacArtney, J. I. <i>et al.</i> (2016) ³⁵	140 (18/22)	not stated	34 cancer, 6 cardiac/lung/renal/liver disease/neurodegenerative disease	not stated	none	not stated	The second half of 'Home is where ...' and 'When a hospital is ...', and all of 'Locating dying as ...'	Dying at home was regarded to be a burden for their family, and the participants desired to protect their families from the burden. However, the struggles existed in complex dynamics and balances.

McPher son, C. J. <i>et al.</i> (2005) ⁸	15 (5/10)	42-78 y/o	advanced cancer	The majority needed considerable assistance and mainly sitting or lying. 3 did self-caring, mobile, and required little assistance.	none	12 spouses living together, 2 daughters not living together, 1 several family members	All	The participants were concerned physical, social, emotional, future, and aftermath burden on their caregivers but also concerned about implications for themselves. Those feelings made the participants try to ease the burden with problem-centred and emotion- centred strategies.
Oeki, M <i>et al.</i> (2020) ³⁶	11 (5/6)	40-89 y/o	advanced cancer	Performance status: 2-4	none	7 partners, 3 children, 1 parents	All	The participants tried to reduce the burden on their families by doing what they could do as much as possible, expressing or suppressing their feelings, thinking positively, ignoring the feeling, and working on with their family members.

ADL: activities of daily living; ALSFR-S: Amyotrophic Lateral Sclerosis Functional Rating Scale; ALS: amyotrophic lateral sclerosis; CA: content analysis; CRF: chronic renal failure; GT: grounded theory; IPA: interpretative phenomenological analysis; NIV: Non-invasive ventilation; PC: palliative care; QOL: Quality of Life; SPB: self-perceived burden; TA: thematic analysis; WHO: World Health Organization

Table 3. Quality appraisal using the CASP checklist

Question	Section A: Are the results valid?					Section B: What are the results?			Section C: Will the results help locally?	
	1. Was there a clear statement of the aims of the research?	2. Is a qualitative methodology appropriate?	3. Was the research design appropriate to address the aims of the research?	4. Was the recruitment strategy appropriate to the aims of the research?	5. Was the data collected in a way that addressed the research issue?	6. Has the relationship between researcher and participants been adequately considered?	7. Have ethical issues been taken into consideration?	8. Was the data analysis sufficiently rigorous?	9. Is there a clear statement of findings?	10. How valuable is the research?
Study										
Ando, H et al. (2019) ²⁹	+	+	+	?	+	±	+	+	+	+
Ashby, M et al. (2005) ³⁰	+	+	±	±	?	?	?	?	±	±
Broom, A et al. (2012) ³¹	+	+	?	+	?	+	?	?	+	±
Foley, G et al. (2016) ³²	?	+	+	?	+	?	±	?	+	±
Foley, G et al. (2014) ³³	?	+	+	?	+	?	±	?	+	±
Gudat, H et al. (2019) ³⁴	+	+	?	+	+	+	+	?	±	±
MacArtney, J. I. et al. (2016) ³⁵	+	+	?	?	?	±	?	?	+	±
McPherson, C. J. et al. (2005) ⁸	+	+	+	?	?	±	?	+	+	+
Oeki, M et al. (2020) ³⁶	+	+	±	?	?	?	+	?	+	+

yes = + higher quality regarding both methodology and reporting

no = - lower quality regarding both methodology and reporting

can't tell = ? inadequate information to make a judgement was provided (insufficient reporting quality)

somewhat = ± relevant data regarding both strengths and limitations were presented but 'to some extent' quality (insufficient methodological quality)²⁸

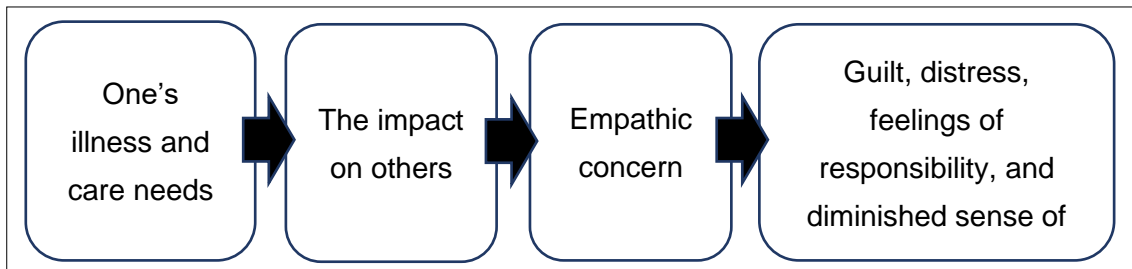


Figure. 1 McPherson's model of self perceived burden⁸

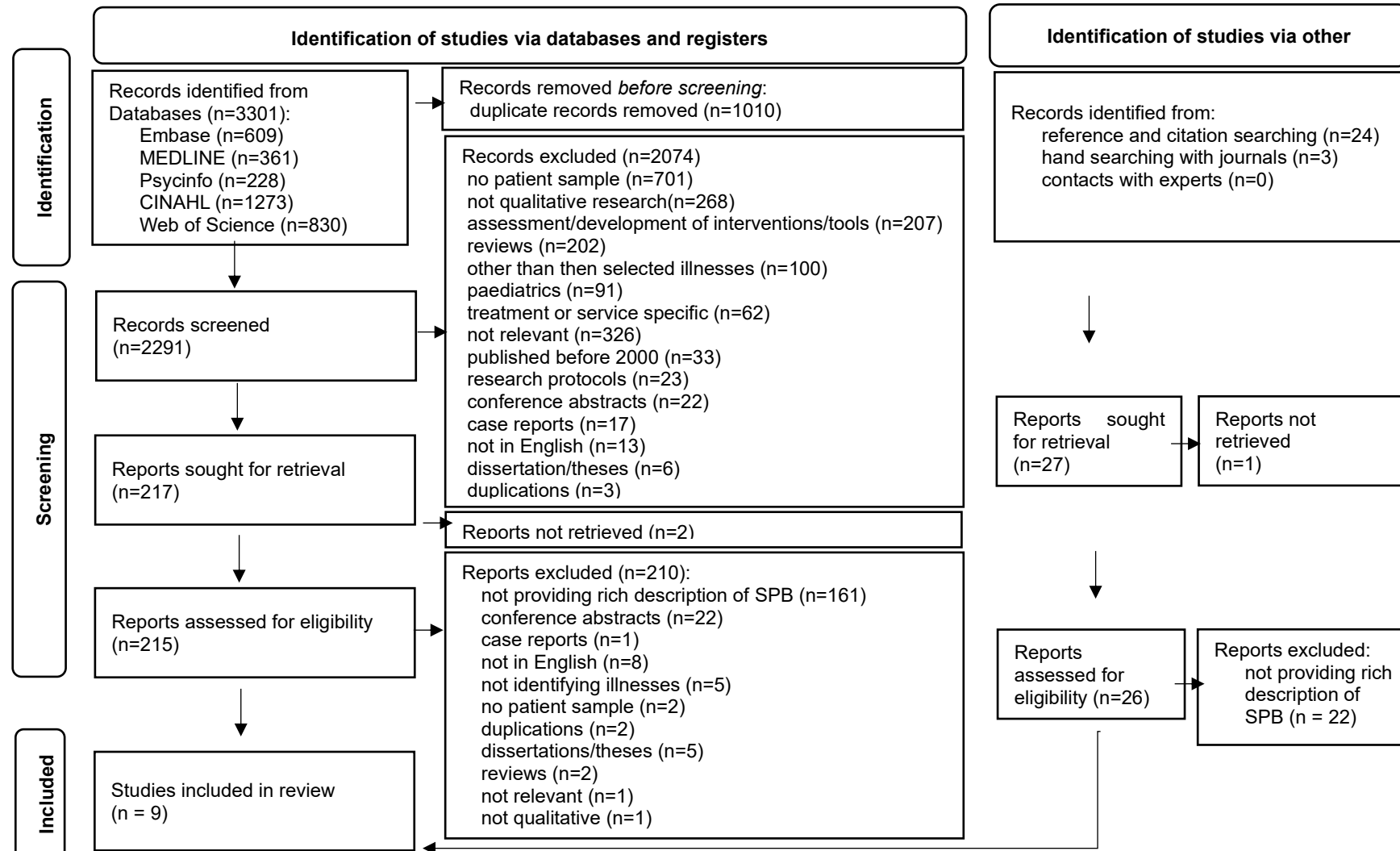


Figure 2. PRISMA 2020 flow diagram

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