Toward the development of a model of caregiver-specific fear of cancer recurrence: a systematic review

Kyra Webb\textsuperscript{a}, Louise Sharpe\textsuperscript{a}, Phyllis Butow\textsuperscript{a,b}, Haryana Dhillon\textsuperscript{a,b,c}, Robert Zachariae\textsuperscript{d,e}, Nina Møller Tauber\textsuperscript{d}, Mia Skytte O'Tool\textsuperscript{d}, Joanne Shaw\textsuperscript{a,b,*}

Abstract

Background: Fear of cancer recurrence (FCR) is the most common psychological issue for cancer survivors, and research shows that caregivers are as fearful of the cancer returning as patients. However, there is relatively little research on caregiver FCR. The aim of this systematic review was to provide a metasynthesis of qualitative research to determine (a) whether caregiver FCR was conceptually similar to FCR among survivors, (b) to determine any specific issues that were different for caregivers compared with survivors, and (c) to present a hypothesis-generating model of caregiver FCR to inform future theoretically grounded caregiver-specific FCR research.

Methods: Using keywords relating to FCR, caregivers, and cancer, CINAHL, PsycINFO, PubMed, and Embase databases were searched. Qualitative studies reporting on FCR in caregivers published between January 1997 and July 2021 were included.

Results: Following PRISMA guidelines, 13 articles were included for review and metasynthesis. Overall, there was a paucity of qualitative research exploring caregiver FCR. Metasynthesis revealed 1 theme, uncertainty/fear previously identified among survivors and a theme related to the caregiver’s experience of FCR: caregiver’s role as protector. An overarching theme, fear of losing a loved one, explained the relationship between each of the identified themes, acting as a driver of caregiver FCR.

Conclusions: This review highlighted that although similarities between survivor and caregiver experiences of FCR exist, key elements that underlie caregiver FCR are conceptually different to the fear experienced by cancer survivors. We propose a new model of FCR that incorporates aspects unique to caregivers which requires further investigation. The proposed model provides an important foundation for further research exploring caregiver FCR. Given caregivers with higher FCR experience more caregiver burden, it is essential to better understand their experiences. This will facilitate the development of interventions which effectively support caregivers, enhancing their capacity to support survivors.

Keywords: Cancer, Caregiver, Family, Fear of cancer recurrence, Oncology, Systematic review

1. Introduction

Early diagnosis and treatment of cancer has advanced significantly, improving outcomes for almost all cancer types\textsuperscript{[1]}; however, the risk of cancer returning or progressing remains a central concern for many survivors.\textsuperscript{[2]} Fear of cancer recurrence (FCR) is currently defined as the “fear, worry or concern relating to the possibility that cancer will come back or progress.”\textsuperscript{[3]} Help managing FCR is identified by cancer survivors as one of the most common unmet needs.\textsuperscript{[4]} Around 50% of survivors experience moderate to high levels of FCR, which is indicative of clinical levels.\textsuperscript{[3,5–7]} Although fears and concerns associated with the uncertain nature of the survivorship trajectory are common,\textsuperscript{[8]} and in some cases adaptive,\textsuperscript{[9]} these fears can become highly debilitating, significantly affecting individual well-being.\textsuperscript{[10]} The experience of FCR has been associated with increased anxiety\textsuperscript{[11]} and psychological distress,\textsuperscript{[12]} lower quality of life,\textsuperscript{[13]} and increased use of health care resources in survivors.\textsuperscript{[9,10]}

For survivors, FCR is characterized by intrusive thoughts which cause excessive distress; maladaptive coping techniques, such as avoidance-based coping\textsuperscript{[14]}; impairments in functioning; and difficulties planning for future events.\textsuperscript{[3,10]} The content of these recurrence fears may be specific in nature, for example, relating to the prospect of receiving a particular type of treatment again, or may be more general, associated with the uncertainty surrounding initial disease prognosis, progression, and/or death.\textsuperscript{[10]} Qualitative data have revealed that survivors with increased FCR report difficulties controlling fear and responding to triggers (e.g., physical symptoms, medical appointments),\textsuperscript{[15,16]} limited use of coping strategies,\textsuperscript{[14]} intolerance of uncertainty,\textsuperscript{[7]} and death-related thoughts.\textsuperscript{[10]}
A number of theoretical frameworks for understanding the development and maintenance of FCR in cancer survivors have been proposed. While these vary in content, there is a consensus about the key elements which underpin FCR. First, cues which may be either internal (eg, treatment side effects) or external (eg, cancer-related information within the media) activate cognitive schemas. Cues are then interpreted using an appraisal process prompting the use of coping strategies which may be more or less beneficial. Initial models proposed that FCR varied depending on one’s illness representation and internal or external cues activating a response. More recent theories have proposed the role of alternate variables such as interpretation bias, metacognitions about worry, and emotional processing. Previously, models focused on content of recurrence fears; however, more recently, a focus on cognitive processes (eg, worry) has been demonstrated. These fears centralize around threats to health, concerns about death and dying, and the implications associated with returning to treatment. Exploration of the content of these fears has greatly enhanced understanding of survivor experiences of FCR and has contributed to the development of a number of successful interventions.

Cancer survivors do not undertake this journey alone. FCR may affect caregivers and survivors. Partners, friends, and other family members often adopt the role of caregiver. These individuals usually share significant, preexisting relationships and play a key role in supporting survivors well after the completion of treatment. Beyond the practical aspects associated with treatment, such as assistance with medication and travel to appointments, caregivers help survivors physically and emotionally. Previous research has suggested the burdens associated with caregiving are substantial. Caregivers have been found to experience similar or even higher levels of distress when compared with survivors. Among distressed caregivers, rates of burnout, poor quality of life, and mental health disorders, particularly depression and anxiety, are high. Sudden role change, in conjunction with cancer-related stressors, may significantly affect patient-caregiver relationships. Caregiver burden is also associated with a range of physical health complications including sleep difficulties and fatigue, cardiovascular disease, poor immune functioning, and increased mortality. Moreover, caregivers engage in caring continguously with other familial obligations which remain constant. Although the initial stages of treatment are associated with considerable disruption to everyday life and great uncertainty, caregiver demands do not cease when treatment ends.

Consistent with findings in survivor populations, caregivers also report FCR as a key concern. In a study examining levels of FCR in survivor-caregiver dyads, a relationship between survivor and caregiver FCR was observed. Caregivers of survivors reporting high levels of FCR also reported similar or higher levels of FCR themselves. Similarly, in caregivers of patients with head and neck cancer, higher levels of FCR were observed in caregivers when compared with survivors. Research that has examined the influence of caregiver FCR on survivor levels of FCR found a reciprocal relationship. These findings indicate that heightened FCR in caregivers increases subsequent FCR in survivors, in turn, leading to an increase in caregiver FCR.

Despite the recognized importance of FCR in caregivers, it is unclear whether the theoretical conceptualizations developed for survivors apply to caregiver FCR, limiting the development of interventions which effectively target caregiver needs. To date, there is only 1 model addressing caregiver FCR. This model emphasizes the role of cancer appraisal by both the survivor and the caregiver as predictors for the development of FCR. The model predicts that both caregivers and patients’ appraisals influence their FCR levels. However, levels of FCR also influence discussions about cancer between survivor-caregiver dyads. When discussing cancer, fearful caregivers avoid speaking about recurrence, reducing cognitive processing for the survivor. In turn, this increases levels of FCR and distress. Conceptually, this model draws heavily on fundamental theories that focus primarily on one’s appraisal of the cancer. Although more recent theories of FCR in survivors have proposed the importance of other variables, such as metacognitive and existential variables, these have not yet been incorporated into a caregiver model of FCR. Developing an appropriate model requires a greater understanding of how caregivers experience FCR and whether these experiences are distinct from those reported by survivors.

To date, only one review has synthesized literature examining FCR in caregivers. This quantitative systematic review (n = 16 studies) found age, treatment type, and psychological beliefs (illness perceptions) predicted FCR. However the review reported inconsistent results for psychological and interpersonal factors, likely because of the small number of appropriately powered studies. The review findings highlight the paucity of research studies and the need for evidence to support theoretical models to better understand FCR in caregivers. A limitation of the review was excluding qualitative studies that may provide greater depth of understanding of the lived experience of caregivers to inform the development of caregiver FCR models. The primary aim of this systematic review, therefore, was to examine the phenomenology of FCR in caregivers, through a metasynthesis of available qualitative studies. Specifically, we aimed to (a) determine whether caregiver experiences of FCR were conceptually similar to those of survivors, (b) explore FCR issues specifically pertaining to caregivers, and (c) present a model of caregiver FCR to provide a foundation for theoretically guided research.

2. Methods

This study was preregistered with PROSPERO international prospective register of systematic reviews (registration number: CRD42020201879), and the results are reported in accordance with the PRISMA guidelines.

2.1. Search strategy

Initial searches were conducted between April and July 2020 of PsychINFO, PubMed, CINAHL, and Embase electronic databases to identify qualitative research examining caregiver experiences of FCR. Searches were updated in June 2021. A search strategy was developed including key words related to cancer (eg, tumor/* cancer*), the population of interest (carer* or caregiver* or informal care or dyad or spouse or relative*), and the concept for investigation (eg, fear* or worry* and recur* or reoccur*). Studies from 1997 onward were included because of the emergence of FCR literature during this period. An example search strategy is included as Appendix A, http://links.lww.com/OR9/A34.

2.2. Inclusion and exclusion criteria

Studies were included for review if they met the inclusion criteria: (a) peer-review publications between January 1997 and June 2021, (b) English language, (c) used qualitative research methods (mixed-methods studies were included if they reported qualitative data separately), (d) sample at least partially comprising cancer caregivers older than 18 years providing care for an adult patient with any cancer type, and (e) reported on caregiver experiences
related to FCR or a related psychological construct, such as anxiety or uncertainty. “Grey literature,” for example, conference abstracts, dissertations, commentaries, opinion pieces, and books were excluded. Studies including caregivers of pediatric patients with cancer were also excluded.

After completion of the searches, title and abstracts were screened independently by 2 authors (K.W. and J.S.). Full-text review was then conducted independently by 2 authors (K.W. and J.S. or L.S.). Reasons for exclusion were recorded (Fig. 1), and any discrepancies were discussed among the review team until agreement was reached.

2.3. Quality appraisal

The Critical Appraisal Skills Programme checklist for qualitative studies was used to evaluate study quality.[55] This consisted of 3 domains covering study validity, result identification, and study applicability. Each section was rated using the following scoring system: 1 = yes; 0 = no; 0 = cannot identify. This rating was completed independently by 2 authors (K.W. and J.S.).

2.4. Extraction and data analysis

Data extraction using a predeveloped template was conducted independently by 2 authors (K.W. and J.S.). Data extracted included (a) study characteristics, including aim and sample size; (b) participant characteristics such as age of caregiver, patient’s cancer type, and relationship to patient; and (c) themes and relevant quotes.

For analysis, we extracted every quotation that referred to fears of cancer recurrence.[56] We intended to analyze the data at the study level and meta-analyze the themes and subthemes. However, FCR was identified as a theme in only 3 studies, and only in 2 of these, were subthemes identified. The remaining studies identified FCR as a subtheme, and hence, they were eligible for inclusion. As a result, we opted to analyze the data at the level of the individual data. Our analysis was guided by Finfgeld-Connett (2018) “A guide to qualitative metasynthesis.”[56] The findings were categorized based on similar experiences and meaning. Initially, similar concepts were grouped using line-by-line coding. A thematic analysis was then conducted to identify common themes across texts and generate new themes based on the caregiver experiences reported. These identified themes were then discussed among the review team and potential additional categories encapsulating these themes identified.[57] Finally, a model was constructed to demonstrate the relationship between themes and provide a basis for further research examining caregiver experiences related to FCR.

3. Results

A total of 5486 studies were identified after database searches, with 3158 remaining after removal of duplicates. Figure 1 summarizes the identification and inclusion of articles. After title and abstract

---

**Figure 1.** PRISMA flow diagram.
screening, a total of 88 articles were reviewed in full, resulting in data being extracted from 13 studies. Reference lists of included articles were also hand-searched returning no further texts for inclusion. Although no studies reported the primary aim of exploring FCR, all identified FCR as an important theme emerging from the analyses.

### 3.1. Study characteristics

In total, qualitative data were analyzed for 260 caregivers. Study characteristics are outlined in Table 1. Relationship with survivor included husband or wife, friend, sibling, child, or parent. Most studies included spouse caregivers,[28,58-63] 3 studies reported a mixed sample,[66-68] and 2 did not specify the relationship.[65,70] Four studies included both survivors and caregivers either as a dyad[28,65] or as a separate unit of analysis.[61,70] Qualitative data were cross-sectional and predominantly collected using semistructured interviews (n = 10).[28,58-61,63,64,66,68,69] Other data collection methodologies included use of a single, open-ended prompt (n = 1),[62] written responses (n = 1),[70] or photovoice in combination with group interviews (n = 1).[65] Patient cancer types included head and neck (n = 2),[59,60] breast (n = 3),[59,61,63] lung (n = 1),[62] ovarian (n = 1),[70] pancreatic (n = 1),[65] and mixed cancer samples (n = 3).[64,66,68] One study identified a specific focus on caregivers providing care for individuals with advanced disease.[66] The average age of caregivers ranged between 31 and 79 years.

Study quality was generally high (Table 2). The 2 components of study quality with the highest average rating were an adequate description of the research setting and methodological approach (Table 2). The 2 components of study quality with the lowest average rating were a discussion of the value of study findings and an adequate description of the relationship between researcher and participants (Table 2). Qualitative data were cross-sectional and predominantly collected using semistructured interviews (n = 10). Other data collection methodologies included use of a single, open-ended prompt (n = 1),[62] written responses (n = 1),[70] or photovoice in combination with group interviews (n = 1).[65] Patient cancer types included head and neck (n = 2),[59,60] breast (n = 3),[59,61,63] lung (n = 1),[62] ovarian (n = 1),[70] pancreatic (n = 1),[65] and mixed cancer samples (n = 3).[64,66,68] One study identified a specific focus on caregivers providing care for individuals with advanced disease.[66] The average age of caregivers ranged between 31 and 79 years.

Study quality was generally high (Table 2). The 2 components of study quality with the highest average rating were an adequate description of the research setting and methodological approach (Table 2). The 2 components of study quality with the lowest average rating were a discussion of the value of study findings and an adequate description of the relationship between researcher and participants (Table 2). Qualitative data were cross-sectional and predominantly collected using semistructured interviews (n = 10). Other data collection methodologies included use of a single, open-ended prompt (n = 1),[62] written responses (n = 1),[70] or photovoice in combination with group interviews (n = 1).[65] Patient cancer types included head and neck (n = 2),[59,60] breast (n = 3),[59,61,63] lung (n = 1),[62] ovarian (n = 1),[70] pancreatic (n = 1),[65] and mixed cancer samples (n = 3).[64,66,68] One study identified a specific focus on caregivers providing care for individuals with advanced disease.[66] The average age of caregivers ranged between 31 and 79 years.

Study quality was generally high (Table 2). The 2 components of study quality with the highest average rating were an adequate description of the research setting and methodological approach (Table 2). The 2 components of study quality with the lowest average rating were a discussion of the value of study findings and an adequate description of the relationship between researcher and participants (Table 2). Qualitative data were cross-sectional and predominantly collected using semistructured interviews (n = 10). Other data collection methodologies included use of a single, open-ended prompt (n = 1),[62] written responses (n = 1),[70] or photovoice in combination with group interviews (n = 1).[65] Patient cancer types included head and neck (n = 2),[59,60] breast (n = 3),[59,61,63] lung (n = 1),[62] ovarian (n = 1),[70] pancreatic (n = 1),[65] and mixed cancer samples (n = 3).[64,66,68] One study identified a specific focus on caregivers providing care for individuals with advanced disease.[66] The average age of caregivers ranged between 31 and 79 years.

Study quality was generally high (Table 2). The 2 components of study quality with the highest average rating were an adequate description of the research setting and methodological approach (Table 2). The 2 components of study quality with the lowest average rating were a discussion of the value of study findings and an adequate description of the relationship between researcher and participants (Table 2). Qualitative data were cross-sectional and predominantly collected using semistructured interviews (n = 10). Other data collection methodologies included use of a single, open-ended prompt (n = 1),[62] written responses (n = 1),[70] or photovoice in combination with group interviews (n = 1).[65] Patient cancer types included head and neck (n = 2),[59,60] breast (n = 3),[59,61,63] lung (n = 1),[62] ovarian (n = 1),[70] pancreatic (n = 1),[65] and mixed cancer samples (n = 3).[64,66,68] One study identified a specific focus on caregivers providing care for individuals with advanced disease.[66] The average age of caregivers ranged between 31 and 79 years.

Study quality was generally high (Table 2). The 2 components of study quality with the highest average rating were an adequate description of the research setting and methodological approach (Table 2). The 2 components of study quality with the lowest average rating were a discussion of the value of study findings and an adequate description of the relationship between researcher and participants (Table 2). Qualitative data were cross-sectional and predominantly collected using semistructured interviews (n = 10). Other data collection methodologies included use of a single, open-ended prompt (n = 1),[62] written responses (n = 1),[70] or photovoice in combination with group interviews (n = 1).[65] Patient cancer types included head and neck (n = 2),[59,60] breast (n = 3),[59,61,63] lung (n = 1),[62] ovarian (n = 1),[70] pancreatic (n = 1),[65] and mixed cancer samples (n = 3).[64,66,68] One study identified a specific focus on caregivers providing care for individuals with advanced disease.[66] The average age of caregivers ranged between 31 and 79 years.

Study quality was generally high (Table 2). The 2 components of study quality with the highest average rating were an adequate description of the research setting and methodological approach (Table 2). The 2 components of study quality with the lowest average rating were a discussion of the value of study findings and an adequate description of the relationship between researcher and participants (Table 2). Qualitative data were cross-sectional and predominantly collected using semistructured interviews (n = 10). Other data collection methodologies included use of a single, open-ended prompt (n = 1),[62] written responses (n = 1),[70] or photovoice in combination with group interviews (n = 1).[65] Patient cancer types included head and neck (n = 2),[59,60] breast (n = 3),[59,61,63] lung (n = 1),[62] ovarian (n = 1),[70] pancreatic (n = 1),[65] and mixed cancer samples (n = 3).[64,66,68] One study identified a specific focus on caregivers providing care for individuals with advanced disease.[66] The average age of caregivers ranged between 31 and 79 years.

Study quality was generally high (Table 2). The 2 components of study quality with the highest average rating were an adequate description of the research setting and methodological approach (Table 2). The 2 components of study quality with the lowest average rating were a discussion of the value of study findings and an adequate description of the relationship between researcher and participants (Table 2). Qualitative data were cross-sectional and predominantly collected using semistructured interviews (n = 10). Other data collection methodologies included use of a single, open-ended prompt (n = 1),[62] written responses (n = 1),[70] or photovoice in combination with group interviews (n = 1).[65] Patient cancer types included head and neck (n = 2),[59,60] breast (n = 3),[59,61,63] lung (n = 1),[62] ovarian (n = 1),[70] pancreatic (n = 1),[65] and mixed cancer samples (n = 3).[64,66,68] One study identified a specific focus on caregivers providing care for individuals with advanced disease.[66] The average age of caregivers ranged between 31 and 79 years.

Study quality was generally high (Table 2). The 2 components of study quality with the highest average rating were an adequate description of the research setting and methodological approach (Table 2). The 2 components of study quality with the lowest average rating were a discussion of the value of study findings and an adequate description of the relationship between researcher and participants (Table 2). Qualitative data were cross-sectional and predominantly collected using semistructured interviews (n = 10). Other data collection methodologies included use of a single, open-ended prompt (n = 1),[62] written responses (n = 1),[70] or photovoice in combination with group interviews (n = 1).[65] Patient cancer types included head and neck (n = 2),[59,60] breast (n = 3),[59,61,63] lung (n = 1),[62] ovarian (n = 1),[70] pancreatic (n = 1),[65] and mixed cancer samples (n = 3).[64,66,68] One study identified a specific focus on caregivers providing care for individuals with advanced disease.[66] The average age of caregivers ranged between 31 and 79 years.
because clinical implications and identification of future research directions were commonly not reported in articles.\(^{55}\)

Synthesis of findings across studies and subsequent qualitative analysis revealed 2 key themes and 2 subthemes which together, centered around 1 overarching theme — fear of losing a loved one. The occurrence of these themes is outlined in Table 3. One theme aligned with survivor experiences of FCR and included uncertainty/fear with 2 subthemes: triggers and protective monitoring. A second theme was identified as caregiver’s role as protector. The relationship between themes is outlined in Fig. 2.

### 3.1.1. Uncertainty/fear

Uncertainty and fear were identified as core concerns across various study settings. This unease was pervasive and centralized around the ongoing fear that cancer may return unannounced at any time:

“No matter what kind of wonderful day you’re having, you know that these black clouds are there and on any day, life could change again in a minute. So you never ever really are without feeling that” [Female, partner].\(^{65}\)

Caregivers felt challenged by the uncertainty about what recurrence may mean, making it difficult to plan for the future:

“And he’s doing really well, but it’s just the fear of the unknown. You never know what can change, how it can come back. Um, just fear of the unknown” [Not specified in primary paper].\(^{62}\)

When thinking of the patient’s cancer reoccurring, caregivers generally expressed 2 core concerns. The stress and unpredictability associated with a recurrence and what this may mean for the patient’s prognosis, combined with an inherent fear of re-experiencing the distress associated with initial diagnosis and treatment. This fear of the future was debilitating and prevented caregivers from moving forward, resulting in a limbo-like state:

“One cannot plan ahead, one continues living as if traumatised, in continuous fear that something bad will happen. The fear that..."
cancer can come back or that there is metastasis to other organs is always there” [Not specified in primary paper].

### 3.1.2. Caregiver’s role as protector

Caregivers expressed a desire to protect their loved one from both external cancer-related stimuli and their own worries and concerns. Caregivers felt a need to shield their loved one from bad news which may cause distress or instil further fears:

“When controlling my emotions so that the focus remains on my sister’s health and wellbeing was fine, as her experience was my number one priority... but now that she has died I am overwhelmed...grief that I have been feeling since the day she was diagnosed that I kept inside to protect her” [Not specified in primary paper].

For some, lack of open communication meant that negative emotions and concerns were internalized, influencing later emotional processing:

“Controlling my emotions so that the focus remains on my sister’s health and wellbeing was fine, as her experience was my number one priority... but now that she has died I am overwhelmed... grief that I have been feeling since the day she was diagnosed that I kept inside to protect her” [Not specified in primary paper].

Subtheme 1: triggers

After a loved one’s cancer diagnosis, caregivers expressed a newfound sensitivity to cancer-related stimuli. This was not situation-specific; instances within the media and personal encounters, such as being informed a relative or friend had been diagnosed with cancer, acted as a stark reminder of their loved one’s diagnosis:

“It’s- since that day everything seems to be cancer. You see it in the papers, you see it on the television and it just comes into the mind again and brings it all up again” [Male, husband].

Constant encounters with cancer-related stimuli facilitated maintenance of recurrence concerns. These worries were ever-present for caregivers, linking to initial experiences of the cancer journey, which were often highly distressing and confusing for both survivors and caregivers, evoking strong emotions:

“I would say that this cancer thing... it does recede a little bit, but it’s always there, and every time anything happens - the whole thing comes back again. And that for me would be absolutely horrific” [Male, 70, Married].

Subtheme 2: protective monitoring

Caregivers also expressed a tendency to watch the survivor closely in a hope that this vigilance would facilitate early detection of changes which may be indicative of a cancer recurrence. This protective monitoring provided a medium where caregivers attempted to assert control over the situation. Caregivers perceived that earlier identification of recurrence would facilitate better prognosis:

“Whenever I see something like, lately the past few days I noticed she’s been like on the verge of cold or something. And she coughs. And every time she coughs, I say, ‘Is that related to the cancer?’” [Not specified in primary paper].

Some caregivers expressed that despite being conducted with positive intentions, this monitoring caused agitation for the survivor:

“I just didn’t want to leave [my loved one]. I was hypervigilant. [I watched] every little thing, then I had to catch myself, you know, and back down because [my loved one] would get annoyed by it... I was] really being a horrible nag about it” [Not specified in primary paper].

### 3.2. Overarching theme: fear of losing a loved one

Driving uncertainty/fear and a desire to protect was the inherent fear of losing a loved one. Consideration of recurrence provoked concern, leading caregivers to evaluate the ongoing threat that cancer poses:
“It is life threatening. You don’t trust cancer. You can’t ignore this elephant that has moved into your life. There’s just no ignoring it” [Not specified in primary paper][59]

When faced with the prospect of losing their loved one, caregivers experienced distress and uncertainty. Caregivers were instead left to consider the worst possible outcome, questions surrounding if, and when this may occur unanswerable:

“Yeah. Just not knowing… it’s like, the whole illness uncertainty of, is she going to live? Is she going to die? And when is she going to die? What am I gonna do after she’s dead? What is the family going to do? What are the last days going to be like?” [Not specified in primary paper][28]

This fear was described as ever-present. The acknowledgment that recurrence may occur and how rapidly this may change everyone’s lives and plans further elevated distress. This led to the consideration of what this would mean both personally and for the wider family:

“I am afraid that she would not respond to chemotherapy and die. After everything we’ve been through, she would die and the family would be completely my responsibility. Nothing else matters” [Not specified in primary paper][58]

This fear was also driven by the uncertainty about both a recurrence and the caregiver’s emotional response to the recurrence:

“The fear of the unknown, the fear of not being sure of how it’s going to happen and how I’m going to react … I’m afraid of losing him … I’m worried about how I’m going to feel…” [Female, Wife][71]

4. Discussion

Since its initial conceptualization, research examining FCR among survivors has increased exponentially.[3,19] Despite the shared experience between survivor and caregiver, the latter have received relatively little attention. This review sought to examine FCR in caregivers and determine how caregiver experiences of FCR are qualitatively different to the experiences reported by survivors. In addition, this review proposes a model of caregiver-specific FCR that incorporates the fear of losing a loved one as a key driver of cancer-related concerns and protective behaviors. These were expressed in our thematic analysis as uncertainty/fear that included 2 subthemes, triggers and protective behaviors. These were expressed by multiple caregivers across studies, indicating relevance.

The ongoing worry associated with the threat of cancer evoked consideration of death.[10] In survivors, identified content of fears have centralised around death and the process of dying.[21,24,72] Given a cancer diagnosis is experienced by survivors and caregivers as a stressful and life-altering event, it is perhaps unsurprising that caregivers also report fears about death.[60] Specifically, we identified uncertainty/fear as a core theme which persisted for years after diagnosis.[60] These were underpinned by the pervasive concern that the survivor may die if recurrence were to occur, driving FCR.[71] This fear was 2-fold: Caregivers also felt to consider life without their loved one and how they themselves may cope.[28,64,73]

To the best of our knowledge, no studies have specifically examined both death anxiety and FCR in caregivers of patients with cancer. Previous research has, however, examined the relationship between quality of life and death anxiety, demonstrating an inverse relationship between these constructs.[74,75] In cancer survivors, fears about death and dying were identified as predictors of FCR,[22] highlighting the importance of investigating the content of recurrence fears.[21] These findings support the relevance of FCR models which derive theoretical elements from existential psychology, particularly terror management theory.[18,22,76] These models are yet to be tested in caregiver populations, and further investigation is needed to determine whether they appropriately capture caregiver experiences of FCR.

Our qualitative systematic review provided unique insight into the constructs that explain how caregivers experience FCR, providing an important foundation for future theoretically grounded research. First, identification of the theme, caregiver’s role as protector, highlights unique concerns specific to this role. This was reported as a desire to protect the survivor from both cancer-related stimuli and their own personal concerns about recurrence. Furthermore, caregivers expressed a reluctance to communicate their own worries or concerns in case it increased patient distress.[58,60] Accordingly, caregivers reported feigning positivity, providing reassurance that may be unrealistic, and shutting down conversations about the realities of recurrence.[60]

Previous quantitative research has explored the role of protective buffering in predicting increased FCR levels in survivor–caregiver dyads.[77] Protective buffering is defined as the hiding or denial of concerns or worries undertaken with the intent of avoiding cancer-related discussions and disagreements.[78] The findings indicated that protective buffering was associated with increased FCR in the individual (patient or partner) engaging in protective buffering.[77] Furthermore, in relationships rated more highly on the relationship satisfaction scale, survivor engagement in protective buffering increased distress levels in partners.[79] This avoidance of conversation around disease recurrence and prognosis, implemented as a protective mechanism,[60] may adversely affect FCR levels given the seemingly reciprocal nature of FCR in survivor–caregiver dyads.[82]

Finally, our review highlights a paucity of literature examining caregiver experiences of FCR. Importantly, FCR was not identified as a primary focus in any of the studies; however, it was a concern expressed by multiple caregivers across studies, indicating relevance. Furthermore, only 1 FCR model to date has included informal caregivers, despite the intrinsic role these individuals play in providing support for survivors.[29,33] Our findings demonstrate similarities between survivor and caregiver experiences of FCR; however, there are distinct elements relating specifically to the ongoing role of caregivers, which current FCR models do not account for. While quantitative studies have incorporated adapted patient measures of FCR for use in caregiver populations,[53,81] the current findings solidify the need for further research examining how caregiver experiences of FCR are characterized.

4.1. Study limitations

Because all studies identified a main focus on topics other than FCR, texts were limited in the depth of content relating specifically to FCR and associated subthemes. In each article, only a limited number of quotes pertaining to caregiver experiences of FCR were
reported, limiting our ability to conduct a metasynthesis at a study level. As a result, we opted for a metasynthesis at the level of individual quotes to facilitate the development of a model to guide future research in this area. Forthcoming studies focusing specifically on FCR and related constructs in caregiver samples will facilitate improved understanding of how caregivers experience FCR. It is also important to note that generalizability of findings is limited because of the lack of sample variance. First, studies were mainly conducted in Western countries meaning experiences may not be reflective of caregivers from different cultural backgrounds. Although the study samples included a variety of cancer types, there was little differentiation in cancer stage, most studies including survivors with nonrecurrent disease and a stage I-III diagnosis. Only 3 of the 13 studies examined caregivers providing care for survivors with advanced disease.[28,58,66] In addition, we limited our search to include only caregivers providing care for adult patients with cancer, and the findings may differ for parents providing care for a child with cancer. Studies were composed of small sample sizes, with 50% including a sample size <10, and predominantly spouses.[28,58–63,65] Although other relationships were included, these were not widely examined, and the experiences of nonsnpose caregivers may differ.

### 4.2. Clinical implications

Our metasynthesis proposes a caregiver-specific FCR model where fear of losing a loved one acts to drive fear and uncertainty and the desire to protect. The potential model suggests models which are composed of small samples sizes, with 50% including a sample size <10, and predominantly spouses.[28,58–63,65] Although other relationships were included, these were not widely examined, and the experiences of nonsnpose caregivers may differ.

#### 4.2.1. Clinical implications

Our metasynthesis proposes a caregiver-specific FCR model where fear of losing a loved one acts to drive fear and uncertainty and the desire to protect. The potential model suggests models which are composed of small samples sizes, with 50% including a sample size <10, and predominantly spouses.[28,58–63,65] Although other relationships were included, these were not widely examined, and the experiences of nonsnpose caregivers may differ.

### Funding

No funds, grants, or other support was received.

### Authors’ contributions

Conceptualization and protocol development: All authors

Literature search: Kyra Webb

Data screening and analysis: Kyra Webb, Joanne Shaw, Louise Sharpe

Initial draft preparation: Kyra Webb

Draft review/editing: All authors

### Conflicts of interest statement

The authors report no conflicts of interest.

Joanne Shaw and Phyllis Butow are IPOS Advisory Board Members and have been recused from reviewing or making decisions for the manuscript.

### References


Given CW. Family caregiving for cancer patients: the state of the literature and a direction for research to link the informal and formal care systems to improve quality and outcomes. Semin Oncol Nurs 2019;35(4):389–394.


Fronczek AE. A phenomenologic study of family caregivers of patients with head and neck cancers. Oncol Nurs Forum 2015;42(6):593–600.


