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Experience of living with cancer and comorbid illness: a qualitative systematic review of 'cancer plus'

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3 **Experience of living with cancer and comorbid illness: a qualitative systematic review of 'cancer plus'**
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ABSTRACT

Introduction: There are an increasing number of people living with and beyond cancer, whose experience is further complicated by additional long term health conditions in the context of an ageing population. The supportive care needs of this growing patient group should be recognised and addressed. There is a need to explore the experience of living with cancer and comorbid illness in order to develop optimal models of patient-centred care. This protocol describes a systematic review which aims to identify the evidence relating to the experience of 'cancer plus' for patients, informal carers and professionals, and to highlight areas where more research is needed.

Methods and Analysis: A systematic review following PRISMA guidance will be undertaken. Databases Medline, Embase, CINAHL, PsycINFO, ASSIA, Sociological Abstracts, Web of Science, SCOPUS, OpenGrey and ProQuest Dissertations and Theses Global will be systematically searched for articles relevant to patient, carer and professional experiences of 'cancer plus'. Two independent reviewers will screen articles for inclusion and evaluate them according to the Critical Appraisal Skills Programme (CASP) tool. A meta-ethnographic approach will be adopted to synthesise the qualitative evidence to offer new theoretical insights into the topic area and for a patient-centred model of care.

Ethics and dissemination: The review does not require formal ethical review as no direct patient contact or patient identifiable data is used. Conduct of the review has been approved internally by the University of Edinburgh Centre for Population Health Sciences Ethics Review Committee. Results of the review will be published in a generalist peer-reviewed journal and presented at a relevant conference in addition to informing subsequent empirical work by the authors on this topic area.

Strengths and limitations of this study:

- Protocol for a systematic review of current literature exploring a highly important topic presenting a current challenge to health care provision
- Using robust methodology following standardised guidelines
- Scope to inform policy and practice and improve patient care
- Systematic review of qualitative empirical evidence only; does not cover quantitative studies or existing systematic reviews but can give important theoretical insights.
- Large evidence base for experience of cancer and experience of multi-morbidity, but little linking the two bodies of evidence, presenting a challenge to the reviewers.

INTRODUCTION

Due to the ageing population and better treatments, there are an increasing number of people living with and beyond cancer (1). The lived experience, and health and social care needs of these individuals are often complicated by the presence of other comorbid chronic illnesses; as many as 78% of cancer patients are living with at least one other condition, the prevalence of which also increase with age (2, 3). Therefore, the support needs of people living with multiple complex conditions are enduring and need to be identified and addressed (4). There is increasing recognition of the role of primary care in the provision of ongoing support to patients particularly after the completion of hospital-based treatment as patients adjust to life post treatment, dealing with ongoing medical issues related to their cancer treatment or other chronic illness (5, 6). This picture becomes more complex when other long term conditions are also being managed, with implications for the coordination of quality care (7) and self-management (8).

Meeting the needs of people living beyond cancer has been identified as a policy priority (9), which informed the development of the National Survivorship Initiative (5) in England and Wales; survivorship also forms part of the remit for Scotland's Better Cancer Care (10). In many cases survivorship initiatives and research target the clinical needs related to long term side effects (11). However, psychosocial support for survivors of cancer is another important consideration and has implications for the role of primary and community care. While valuable research has been conducted to understand better these dimensions of life for the patient, and their relatives, in addition to the role of and impact on primary care e.g. (12-16), less is known about the impact of additional chronic illness on patients' lived experience. As such, service development and provision would benefit from further in-depth research in this area (6, 17). Understanding the challenges to identity and experiences of people living after a cancer diagnosis with other chronic conditions such as COPD, diabetes or mental ill health, can give new insights into patient-centred models of care; with scope to explore the concept of 'survivorship' in this context.

Objectives

While the holistic needs of cancer survivors form part of the vision of survivorship initiatives, and there is a growing body of work exploring experience of multi-morbidity (8), there is a dearth of research linking cancer with comorbid disease to further enrich our understanding of living with 'cancer plus'. This systematic review aims to synthesise and report on published evidence exploring the experience of cancer survivorship in connection with that of other long term conditions, in particular looking at issues of psycho-social support needs, identity, quality of life, accessing services and quality of care. The findings from the review will iteratively combine with an ongoing patient-centred research prioritisation exercise to map out an empirical research agenda exploring the experience of living with cancer and comorbid illness.

Research questions:

- What are the findings of qualitative evidence exploring the *experience* of living with both cancer and one or more co-morbidities, including personal identity/identities?
- What evidence is there for informal carer and professional perspectives in supporting people with cancer and one or more co-morbidities?
- What are the psychosocial support needs of people living with cancer and other long term condition(s) identified in the literature?
- What are people's experiences of services and quality and coordination of care reported in the literature?
- What research priorities can be derived from the available evidence?

REVIEW METHODS

Design

A systematic review of the current evidence linking experience of cancer with comorbid illness is considered a robust way of identifying and synthesising the published evidence in this area to define a cohesive empirical research agenda that builds on prior knowledge (18). The review will include qualitative evidence

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3 only since the focus is on lived experience and because of the exploratory nature of the topic. It is based on
4
5 the PRISMA statement guidance for conducting a systematic review (19), and the protocol follows the
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7 relevant items on the PRISMA-P checklist (20). The review protocol has been registered on the International
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9 Prospective Register of Systematic Reviews (PROSPERO) database (registration number: CRD42016041796).

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12 The review will use methods of qualitative synthesis to combine, integrate and interpret the evidence from
13
14 the included papers (see eligibility and data synthesis sections below) (21, 22). The review aims to move
15
16 beyond the aggregation of available data to a higher level of interpretation in order to inform the current
17
18 theory around living with complex illness and define where further research can add to what is known (21).

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21 No formal ethical approval is required for this study as it will not include patient identifiable data. It has been
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23 internally approved by the University of Edinburgh Centre for Population Health Sciences Ethics Review
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25 Committee (Ref: 8/03/2016).

26 27 28 *Eligibility criteria*

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30 The review will include qualitative empirical studies, including unpublished grey literature. As above,
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32 quantitative studies will not be included as the review seeks in-depth data exploring lived experiences.
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34 However, qualitative data from mixed methods studies will be screened for inclusion and included if the
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36 qualitative component is relevant. Included articles will be published between 2000 and the present day to
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38 ensure the currency of the work while enabling a broad view of developing issues to be identified. Articles
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40 will be included that address the lived experience of the cancer journey with attention to existing long term
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42 health conditions, and will identify issues related to psychosocial and supportive care as outlined in relation
43
44 to the anticipated dimensions of interest. Articles will be included from across the cancer continuum from
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46 diagnosis through to end of life and will include the perspectives of adult patients (aged 18 or over), informal
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48 carers and health care professionals. No language restriction will be imposed and translations will be sought
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50 for non-English articles.
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55 Studies will be included for any cancer type in combination with one or more comorbid long term condition
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57 (LTC), also described as cancer multi-morbidity or 'cancer plus'. Long term conditions to be included will be
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3 guided by, but not exclusively limited to, those listed in Barnett et al's paper mapping the epidemiology of
4 multi-morbidity (3) (See Appendix 1 for a full list of conditions). The applied definition of long term
5 conditions is based on ISD Scotland's report on important long term conditions in Scotland; the report uses,
6 among others, the Long Term Conditions Collaborative to define an LTC as, 'one that requires ongoing care,
7 limits what one can do and is likely to last longer than a year'(23). Long term side effects of cancer treatment
8 and second primary cancers are not included; experience of second primary cancers is being addressed
9 separately within the research centre. As an exploratory review, a low threshold will be adopted for included
10 conditions as perceived by the individual, but there will be a focus on those that, alongside effects from the
11 cancer and its treatment, are most prevalent and have the greatest impact on individuals' lives (24).
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22 *Dimensions of interest*

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24 The topics covered are expected to include quality of life, experiences of and access to services, supportive
25 care needs, attitude and identity, continuity and perceived quality of care, influence of personal
26 demographics (age, gender, ethnicity, socio-economic status), coordination of care for multiple conditions
27 and experiences of palliative care. However, the outcomes will depend on the evidence available and gaps in
28 the evidence will be highlighted for future study.
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36 *Information sources*

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38 A variety of information sources will be examined to identify potential literature for the review. This will
39 primarily involve a literature database search (including grey literature), but will also include citation and
40 snowball searching, known expert consultation via email, related articles searches in PubMed and use of
41 Google scholar. The databases to be consulted are: Medline, Embase, CINAHL, PsycINFO, ASSIA, Sociological
42 Abstracts, Web of Science, SCOPUS and, for grey literature, OpenGrey and ProQuest Dissertations and
43 Theses Global. These are considered sufficient to comprehensively cover the range of topics and disciplines
44 implicated in this review.
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Search strategy

The searching phase aims to produce a sampling frame from which relevant papers can be selected. The process of searching to be undertaken will begin with individually searching each of the listed databases using an adapted list of search terms; the developed search strategy for Medline is shown in Box 1. The search terms will be adapted to suit each database to derive the most meaningful search and will use free text, MeSH and subject headings for maximum sensitivity and specificity.

Box 1: Example search strategy for Medline

1. Comorbidity/
2. (Multimorbid\$ or multi-morbid\$ or Co-morbid\$).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
3. Chronic Disease/ or (Chronic adj5 (illness\$ or condition\$ or disease)).tw.
4. (Long term adj5 (condition\$ or illness\$ or disease\$)).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
5. 1 or 2 or 3 or 4
6. Neoplasms/ or Cancer.mp or (Tumour or tumor or Oncology or Neoplasm).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
7. Qualitative Research/ or Qualitative.mp or Interview/ r Interview\$.mp or In-depth.mp
8. (Focus Group or Survey or Questionnaire\$ or Ethnograph\$ or Observation\$ or Participant\$ or Respondent\$ or View\$ or Belief\$ or Attitude\$ or Awareness or Perspective\$ or Understanding\$ or Findings or Grounded Theory or Social Construction\$ or Theoretical or Phenomenolog\$).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
9. 7 or 8
10. Psychosocial.mp. or Stress, Psychological/ or Social Support/ or Psycho-social.mp or "Quality of Life"/ or "Experience of Illness".mp or Experience adj5 Illness or Attitude to Health/ or "Quality of Life".mp or Happ\$.mp or Emotion\$.mp. or Emotions/
11. Patient satisfaction.mp. or Patient Satisfaction/ or "Quality of Health Care"/ or "Patient Experience".mp or Satisfaction.mp or "Information Preferences".mp or Decision Making/ or (Decision-making or Decision Making).mp or (Informed choice or Informed Decision Making).mp or Self management.mp. or Self Care/ or "Continuity of Patient Care"/ or Integrated care.mp
12. Access to Health Care.mp. or Health Services Accessibility/
13. Identity.mp. or gender.mp. or Gender Identity/
14. (Health behaviour or Health behavior).mp.
15. (Help-seeking or Help seeking).mp.
16. (Well-being or Well being).mp. or Holistic.mp or Continuity of Care.mp or "Continuity of Patient Care"/ or (Fragmented adj5 care).mp or (joined-up or Joined Up).mp or Integrated care.mp or Diversity of Care.mp.
17. Primary care.mp. or Primary Health Care/ or Secondary care.mp. or Secondary Care/ or Community Care.mp or Community Health Services/ or Ambulatory Care.mp. or Ambulatory Care/ or "Delivery of Health Care, Integrated"/ or Preventive Health Services/ or Family Practice/ or Shared care.mp or Collaborat\$ care.mp

18. (Patient-centred care or Patient centred care or Patient-centred or Person-centred or Patient centred or Person centred).mp or Patient-Centered Care/

19. Palliative Care/ or Supportive care.mp. or palliative.mp.

20. "Health Services Needs and Demand"/ or unmet need.mp. or Health Services Research/ or Depriv\$.mp. or (Socio-economic or Socioeconomic).mp. or Information needs.mp.

21. Survivor\$.mp. or Survivors/

22. Patient Care Planning/ or Care plan.mp. or (Follow-up or Follow up).mp.

23. ("use of service\$" or "service use" or "service adj5 use").mp.

24. 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23

25. 5 AND 6 AND 9 AND 24

DATA COLLECTION AND ANALYSIS

Study records

Identified records will be imported into and managed in EndNoteX7. Screened and selected articles will be managed in subsequent EndNote databases in order to track and record the number of records retained at each step. It is anticipated that given the relative lack of attention in the literature specifically examining the experience of managing long term conditions as a cancer survivor, the review will produce a small number of relevant studies.

Screening

Screening of articles will adhere to the specified inclusion criteria (as detailed above; also see Box 2).

Box 2: Inclusion and exclusion criteria

Inclusion criteria:

- Study population – Adults (over 18) with a diagnosis of cancer and at least one other LTC (as specified in appendix 1), carers and health professionals
- Articles relating to experience of illness from patient, carer and professional perspectives
- Articles focusing on psychosocial and supportive care, identity, quality of life, access to services and quality of care
- Studies with a qualitative empirical design

Exclusion criteria:

- Articles not meeting the above inclusion criterion.
- Articles published before 2000
- Articles focusing on long term side effects of cancer treatment or second primary cancers alone.

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11 A three-step screening process will be undertaken to achieve the final set of included articles. Firstly, all titles
12 will be read for relevance and fit with the inclusion criteria. Articles of no obvious relevance to the subject of
13 the review or referring to the wrong study type (e.g. quantitative or theoretical pieces), will be filtered out at
14 this stage. This process will be undertaken by the primary reviewer DC. Secondly, abstract screening will be
15 undertaken by DC and another reviewer in adherence to systematic review guidelines to look in more detail
16 for relevance and fit with the inclusion criteria in articles where it was not pertinently obvious from the title.
17 Articles deemed irrelevant or of ineligible study type will be filtered out at this stage and included articles
18 highlighted and saved in a new EndNote database. Any differences will be resolved by a third reviewer.
19 Finally, full text articles of included studies will be obtained and read, again by DC and one other reviewer, to
20 assess their suitability for inclusion in the final review. Full text articles deemed to meet the inclusion criteria
21 will be selected and subject to quality appraisal and data extraction.
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36 *Data extraction*

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38 Data extraction will be managed in Microsoft Excel using a purpose designed proforma. Extracted material
39 will be in line with the inclusion criteria and the designated aims of the review, derived from the article as a
40 whole. Information will be gathered on: author; year of publication; country of study; study type; setting;
41 relevant background and impetus for the study; methodological approach and specified methods; main
42 findings including pertinent themes relating to psychosocial needs and supportive care; strengths and
43 limitations; and key relevant discussion points. Extraction of data will be carried out by DC and checked by
44 the second reviewer.
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53 *Quality assessment*

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3 All included articles will be subject to critical appraisal using the Critical Appraisal Skills Programme (CASP)
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5 tool. The 10-item CASP tool was considered to be the most suitable tool to consider the quality parameters
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7 of qualitative work and is a well validated and accepted tool. In reference to the debate on exclusion of
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9 qualitative articles on the basis of quality, no articles will be excluded on these grounds alone and a low
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11 threshold for inclusion will be applied so that the review can benefit from researcher insight and theoretical
12
13 as well as empirical contributions (25-27). The relative quality of included studies will be critically considered
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15 during the analysis and in reference to the developed synthesis. Quality assessment will be undertaken by
16
17 two independent reviewers and any differences of opinion will be discussed and reviewed by a third person
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19 if no consensus is reached.
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22 23 *Data synthesis*

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25 For a qualitative review seeking to explore personal lived experiences, it was important to find a method
26
27 that acknowledges the different paradigms and positions upon which each research output is based as well
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29 as allowing for different study methods covering a broad and exploratory topic area. Meta-ethnography was
30
31 considered an appropriate method for this purpose (22), suited to reviewing a small body of exclusively
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33 qualitative evidence (28). The meta-ethnographic technique was developed and used originally in education
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35 research by Noblit and Hare but has more recently been of interest in nursing and health services research
36
37 (28, 29). Meta-ethnography gives rise to an interpretive account of the descriptive form found in qualitative
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39 research rather than simply aggregating the data. The researcher is encouraged to consider the concepts
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41 and themes that relate different studies and provide a mutual translation to ultimately produce a third order
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43 account of the phenomenon of interest, thus inferring the social and cultural meaning of events (30), as a
44
45 grounded theory approach aims to do with primary data analysis (31, 32).
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50 The meta-ethnographic approach offers three main techniques that will be applied to the extracted data and
51
52 themes: reciprocal translational analysis (RTA); refutational synthesis and lines of argument synthesis (LOA).
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54 RTA will compare themes across the included studies for similarities using the matrix of the themes and
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56 translate and integrate them with an overarching explanatory concept in a similar way to the constant
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3 comparison technique used in a grounded theory approach. Refutational synthesis will identify and explain
4
5 any contradictions across the studies in the same way as deviant cases are utilised to reinforce dominant
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7 concepts. Finally, a LOA synthesis will further develop concepts identified in the RTA to arrive at a 'third
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9 order' or 'synthetic' theoretical framework that synthesises the evidence and provides new insights into the
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11 focus of the review (28).
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13 14 15 **DISCUSSION**

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17 Insights from the proposed review will contribute to what is known, giving novel attention to the
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19 combination of cancer with other long term chronic conditions. This is considered a necessary step in cancer
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21 survivorship research and care in the face of the increasing, ageing population and the changing face of
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23 primary care.
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26 Building on the knowledge base will give rise to an evidence-based and theoretically informed integrated
27
28 model of care, with an emphasis on better understanding experiences of complex and multiple conditions
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30 and the implications for managing 'cancer plus' in the push towards shared care or self-management (33).
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33 The findings from the review will combine with empirical work by the authors to explore the needs of the
34
35 defined patient group to inform the development of a primary care or community based intervention to
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37 better address their complex needs. The review will be published in a peer-reviewed journal and presented
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39 at a relevant conference in addition to being shared with local interest groups and via online social media.
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47
48 literature databases and to Dr Karen Barnett for advice on defining included long term conditions.
49

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52
53 the review protocol with input from CC, SCB, EW and EB. DC is guarantor of the review.
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Appendix 1: List of Long Term Conditions

Hypertension
Depression
Painful condition
Asthma
Coronary Heart Disease
Dyspepsia
Diabetes
Thyroid Disorders
Rheumatoid arthritis, other inflammatory polyarthropathies and systemic connective tissue disorders
Hearing loss
Chronic Obstructive Pulmonary Disease
Anxiety and other neurotic, stress-related and somatoform disorders
Irritable Bowel Syndrome
Cancer
Alcohol Problems
Other psychoactive substance misuse
Constipation
Stroke or transient ischaemic attack
Chronic kidney disease
Diverticular disease of intestine
Atrial fibrillation
Peripheral vascular disease
Heart failure
Prostate disorders
Glaucoma
Epilepsy
Dementia
Schizophrenia
Psoriasis or eczema
Inflammatory bowel disease
Migraine
Blindness and low vision
Chronic sinusitis
Learning disability
Anorexia or bulimia
Bronchiectasis
Parkinson's disease
Multiple Sclerosis
Viral hepatitis
Chronic liver disease

Source: Adapted from: Barnett K, Mercer SW, Norbury M, Watt G, Wyke S, Guthrie B. Epidemiology of multimorbidity and implications for health care, research, and medical education: a cross-sectional study. *Lancet*, 2012;380(9836):37-43.

PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) 2015 checklist: recommended items to address in a systematic review protocol*

Section and topic	Item No	Checklist item
ADMINISTRATIVE INFORMATION		
Title:		
Identification	1a	Identify the report as a protocol of a systematic review
Update	1b	If the protocol is for an update of a previous systematic review, identify as such
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number
Authors:		
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review
Amendments	4	If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments
Support:		
Sources	5a	Indicate sources of financial or other support for the review
Sponsor	5b	Provide name for the review funder and/or sponsor
Role of sponsor or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol
INTRODUCTION		
Rationale	6	Describe the rationale for the review in the context of what is already known
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)
METHODS		
Eligibility criteria	8	Specify the study characteristics (such as PICO, study design, setting, time frame) and report characteristics (such as years considered, language, publication status) to be used as criteria for eligibility for the review
Information sources	9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other grey literature sources) with planned dates of coverage
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated
Study records:		
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review

Selection process	11b	State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (that is, screening, eligibility and inclusion in meta-analysis)
Data collection process	11c	Describe planned method of extracting data from reports (such as piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators
Data items	12	List and define all variables for which data will be sought (such as PICO items, funding sources), any pre-planned data assumptions and simplifications
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale
Risk of bias in individual studies	14	Describe anticipated methods for assessing risk of bias of individual studies, including whether this will be done at the outcome or study level, or both; state how this information will be used in data synthesis
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised
	15b	If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data and methods of combining data from studies, including any planned exploration of consistency (such as I^2 , Kendall's τ)
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies)
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE)

*** It is strongly recommended that this checklist be read in conjunction with the PRISMA-P Explanation and Elaboration (cite when available) for important clarification on the items. Amendments to a review protocol should be tracked and dated. The copyright for PRISMA-P (including checklist) is held by the PRISMA-P Group and is distributed under a Creative Commons Attribution Licence 4.0.**

From: Shamseer L, Moher D, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart L, PRISMA-P Group. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. BMJ. 2015 Jan 2;349(jan02 1):g7647.

BMJ Open

Experience of living with cancer and comorbid illness: protocol for a qualitative systematic review

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Primary Subject Heading:	Oncology
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3 **Experience of living with cancer and comorbid illness: protocol for a qualitative systematic review**
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10 Debbie Cavers, Sarah Cunningham-Burley, Eila Watson, Elspeth Banks, Christine Campbell
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ABSTRACT

Introduction: There is an increasing number of people living with and beyond cancer, whose experience is further complicated by additional long term health conditions in the context of an ageing population. The supportive care needs of this growing patient group should be recognised and addressed. There is a need to explore the experience of living with cancer and comorbid illness in order to develop optimal models of patient-centred care. This protocol describes a systematic review which aims to identify the qualitative evidence relating to the experience of cancer and comorbid illness for patients, informal carers and professionals, and to highlight areas where more research is needed.

Methods and Analysis: A systematic review following PRISMA guidance will be undertaken. Medline, Embase, CINAHL, PsycINFO, ASSIA, Sociological Abstracts, Web of Science, SCOPUS, OpenGrey and ProQuest Dissertations and Theses Global databases will be systematically searched for articles relevant to patient, carer and professional experiences. Two independent reviewers will screen articles for inclusion and evaluate them according to the Critical Appraisal Skills Programme (CASP) tool. Extracted data will be combined using recognised methods of qualitative synthesis to offer new insights into the topic area and for a patient-centred model of care.

Ethics and dissemination: The review does not require formal ethical review as no direct patient contact or patient identifiable data is used. Conduct of the review has been approved internally by the University of Edinburgh Centre for Population Health Sciences Ethics Review Committee. Results of the review will be published in a generalist peer-reviewed journal and presented at a relevant conference in addition to informing subsequent empirical work by the authors on this topic area.

Strengths and limitations of this study:

- Protocol for a systematic review of current literature exploring a highly important topic presenting a current challenge to health care provision
- Using robust methodology following standardised guidelines
- Scope to inform policy and practice and improve patient care
- Systematic review of qualitative empirical evidence only; does not cover quantitative studies or existing systematic reviews but can give important insights.
- Large evidence base for experience of cancer and experience of multi-morbidity, but little linking the two bodies of evidence, presenting a challenge to the reviewers.

INTRODUCTION

Due to the ageing population and better screening, diagnosis and treatments, there are an increasing number of people living with and beyond cancer (1). The lived experience of these individuals is often complicated by the presence of other comorbid chronic illnesses; as many as 78% of cancer patients are living with at least one other condition, the prevalence of which also increase with age (2, 3). Therefore, the support needs of people living with multiple complex conditions are enduring and need to be identified and addressed (4). In addition to the challenges facing secondary care (5), there is increasing recognition of the role of primary care in the provision of ongoing support to cancer patients, particularly after the completion of hospital-based treatment as patients adjust to life post treatment, living with ongoing medical issues related to their cancer treatment (6, 7). This picture becomes more complex in the presence of other long term conditions, with implications for the coordination of quality care and support (8, 9).

Meeting the needs of people living beyond cancer has been identified as a policy priority (10), which informed the development of the National Cancer Survivorship Initiative (6) in England and Wales; survivorship also forms part of the remit for Scotland's Better Cancer Care (11). In many cases survivorship initiatives and research target the clinical needs related to long term side effects (12). However, psychosocial support for survivors of cancer is another important consideration and has implications for the role of primary, secondary and community care. While valuable research has been conducted to understand better these dimensions of life for the patient, and their relatives (13-17), less is known about the impact of additional chronic illness on patients' lived experience. As such, service development and provision would benefit from further in-depth research in this area (7, 18). Understanding the challenges experienced by people living after a cancer diagnosis with other chronic conditions such as COPD, diabetes or mental ill health, can give new insights into patient-centred models of care.

Objectives

While addressing the holistic needs of cancer survivors form part of the vision of survivorship initiatives, and there is growing body of work exploring experience of multi-morbidity (19), there is a dearth of research

1
2
3 linking cancer with comorbid disease to further enrich our understanding of living with such complex illness.
4
5 This systematic review aims to synthesise and report on published evidence exploring the experience of
6
7 cancer survivorship in connection with that of other long term conditions in order to identify critical research
8
9 questions. The findings from the review will combine with those from a separate patient-centred research
10
11 prioritisation exercise to set a qualitative empirical research agenda for future work.
12
13

14 *Research questions:*

- 15
- 16
- 17 • What are the findings of qualitative evidence exploring the *experience* of living with both cancer and
- 18 one or more comorbidities from patient, carer and provider perspectives?
- 19
- 20
- 21 • What are the psychosocial support needs of people living with cancer and one or more other long
- 22 term condition(s) identified in the literature?
- 23
- 24
- 25 • What are patient, carer and provider experiences of service provision reported in the literature?
- 26
- 27
- 28 • What research priorities can be derived from the available evidence?
- 29
- 30

31 **REVIEW METHODS**

32
33
34 *Design*

35
36 A systematic review of the current evidence linking experience of cancer with comorbid illness is considered
37
38 a robust way of identifying and synthesising the published evidence in this area to define a cohesive
39
40 empirical research agenda that builds on prior knowledge (20). The review will include qualitative evidence
41
42 only to produce an interpretation built on people's views and experiences; acknowledging the rich context
43
44 and different dimensions of the lived experience from the perspective of those experiencing it. Further, a
45
46 synthesis of qualitative data aims to generate findings that are meaningful, relevant and appropriate to
47
48 individuals, to inform a qualitative research agenda, and ultimately to more effectively influence policy and
49
50 practice influencing patient outcomes (21).
51
52

53
54 The review is based on the PRISMA statement guidance for conducting a systematic review (22), and the
55
56 protocol follows the relevant items on the PRISMA-P checklist (23). The review protocol has been registered
57
58

1
2
3 on the International Prospective Register of Systematic Reviews (PROSPERO) database (registration number:
4
5 CRD42016041796).

6
7
8 The review will use methods of qualitative synthesis to combine, integrate and interpret, where possible, the
9
10 evidence from the included papers (see eligibility and data synthesis sections below)(21, 24). The review
11
12 aims to move beyond the aggregation of available data to provide further interpretive insights into living
13
14 with complex illness and define where future research can add to what is known (24).
15

16
17 No formal ethical approval is required for this study as it will not include patient identifiable data. It has been
18
19 internally approved by the University of Edinburgh Centre for Population Health Sciences Ethics Review
20
21 Committee (Ref: 8/03/2016).
22

23 24 *Eligibility criteria*

25
26 The review will include qualitative empirical studies, including unpublished grey literature. Qualitative data
27
28 from mixed methods studies will be screened for inclusion and included if the qualitative component is
29
30 relevant. Included articles will be published between 2000 and the present day to ensure the currency of the
31
32 work while enabling a broad view of developing issues to be identified. Articles will be included that address
33
34 the lived experience of the cancer journey with attention to existing long term health conditions (developed
35
36 before or after the cancer diagnosis), and will identify issues related to psychosocial and supportive care as
37
38 outlined in relation to the anticipated dimensions of interest. Articles will be included from across the cancer
39
40 continuum from diagnosis through to end of life and will include the perspectives of adult patients (aged 18
41
42 or over), informal carers and health care professionals. The review will include only articles published in
43
44 English.
45
46

47
48 Studies will be included for any cancer type in combination with one or more comorbid long term condition
49
50 (LTC), also described as cancer multi-morbidity. Long term conditions to be included will be guided by those
51
52 listed in Barnett et al's paper mapping the epidemiology of multi-morbidity (3) (See Appendix 1 for a full list
53
54 of conditions). The applied definition of long term conditions is based on ISD Scotland's report on important
55
56 long term conditions in Scotland; the report uses, among others, the Long Term Conditions Collaborative to
57
58

1
2
3 define an LTC as, 'one that requires ongoing care, limits what one can do and is likely to last longer than a
4
5 year'(25). Long term side effects of cancer treatment and second primary cancers are not included;
6
7 experience of second primary cancers is being addressed separately within the research centre. Where it is
8
9 not possible to determine whether the condition is caused by cancer treatment, the article will be included
10
11 in the review but analysed separately.
12

13 *Dimensions of interest*

14
15 The review focuses on an exploration of the dimensions of lived experience: physical, social, emotional and
16
17 psychological. From a similar study exploring multiple dimensions of experience, these are expected to
18
19 include such topics as quality of life, well-being, psychosocial supportive care needs, attitude and identity,
20
21 perceived continuity and quality of care, and influence of personal demographics (age, gender, ethnicity,
22
23 socio-economic status) (26). In relation to how services can best meet patients' needs, topics may include
24
25 issues of accessing appropriate services, coordination of care for multiple conditions, and self-management
26
27 across the cancer care continuum (17, 19) . The search strategy has been developed to reflect these broad
28
29 and exploratory domains. The outcomes will depend on the evidence available and gaps in the evidence will
30
31 be highlighted for future study.
32
33
34

35 *Information sources*

36
37 A variety of search strategies will be used to identify potential literature for the review, particularly given
38
39 the challenges in identifying qualitative evidence (27). This will primarily involve a literature database search
40
41 (including grey literature), but will also include citation and snowball searching, known expert consultation
42
43 via email, related articles searches in PubMed and use of Google scholar. The databases to be consulted are:
44
45 Medline, Embase, CINAHL, PsycINFO, ASSIA, Sociological Abstracts, Web of Science, SCOPUS and, for grey
46
47 literature, OpenGrey and ProQuest Dissertations and Theses Global. These are considered sufficient to
48
49 comprehensively cover the range of topics and disciplines implicated in this review.
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Search strategy

The searching phase aims to identify relevant studies for inclusion in the review. The process of searching will begin with individually searching each of the listed databases using an adapted list of search terms; the developed search strategy for Medline is shown in Box 1. The search terms will be adapted to suit each database to derive the most meaningful search and will use free text, MeSH and subject headings for maximum sensitivity and specificity.

Box 1: Example search strategy for Medline

1. Comorbidity/
2. (Multimorbid\$ or multi-morbid\$ or Co-morbid\$).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
3. Chronic Disease/ or (Chronic adj5 (illness\$ or condition\$ or disease)).tw.
4. (Long term adj5 (condition\$ or illness\$ or disease\$)).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
5. 1 or 2 or 3 or 4
6. Neoplasms/ or Cancer.mp or (Tumour or tumor or Oncology or Neoplasm).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
7. Qualitative Research/ or Qualitative.mp or Interview/ r Interview\$.mp or In-depth.mp
8. (Focus Group or or Ethnograph\$ or Observation\$ or Participant\$ or Respondent\$ or View\$ or Belief\$ or Attitude\$ or Awareness or Perspective\$ or Understanding\$ or Findings or Grounded Theory or Social Construction\$ or Theoretical or Phenomenolog\$).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
9. 7 or 8
10. Psychosocial.mp. or Stress, Psychological/ or Social Support/ or Psycho-social.mp or "Quality of Life"/ or "Experience of Illness".mp or Experience adj5 Illness or Attitude to Health/ or "Quality of Life".mp or Happ\$.mp or Emotion\$.mp. or Emotions/
11. Patient satisfaction.mp. or Patient Satisfaction/ or "Quality of Health Care"/ or "Patient Experience".mp or Satisfaction.mp or "Information Preferences".mp or Decision Making/ or (Decision-making or Decision Making).mp or (Informed choice or Informed Decision Making).mp or Self management.mp. or Self Care/ or "Continuity of Patient Care"/ or Integrated care.mp
12. Access to Health Care.mp. or Health Services Accessibility/
13. Identity.mp. or gender.mp. or Gender Identity/
14. (Health behaviour or Health behavior).mp.
15. (Help-seeking or Help seeking).mp.
16. (Well-being or Well being).mp. or Holistic.mp or Continuity of Care.mp or "Continuity of Patient Care"/ or (Fragmented adj5 care).mp or (joined-up or Joined Up).mp or Integrated care.mp or Diversity of Care.mp.
17. Primary care.mp. or Primary Health Care/ or Secondary care.mp. or Secondary Care/ or Community Care.mp or Community Health Services/ or Ambulatory Care.mp. or Ambulatory Care/ or "Delivery of Health Care, Integrated"/ or Preventive Health Services/ or Family Practice/ or Shared care.mp or Collaborat\$ care.mp

1
2
3
4 18. (Patient-centred care or Patient centred care or Patient-centred or Person-centred or Patient centred or Person
5 centred).mp or Patient-Centered Care/

6 19. Palliative Care/ or Supportive care.mp. or palliative.mp.

7
8 20. "Health Services Needs and Demand"/ or unmet need.mp. or Health Services Research/ or Depriv\$.mp. or (Socio-
9 economic or Socioeconomic).mp. or Information needs.mp.

10 21. Survivor\$.mp. or Survivors/

11 22. Patient Care Planning/ or Care plan.mp. or (Follow-up or Follow up).mp.

12 23. ("use of service\$" or "service use" or "service adj5 use").mp.

13 24. 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23

14 25. 5 AND 6 AND 9 AND 24
15
16

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19
20 **DATA COLLECTION AND ANALYSIS**

21 *Study records*

22
23 Identified records will be imported into and managed in EndNoteX7. Screened and selected articles will be
24 managed in subsequent EndNote databases in order to track and record the number of records retained at
25 each step. It is anticipated that given the relative lack of attention in the literature specifically examining the
26 experience of long term conditions as a cancer survivor, the review will produce a small number of relevant
27 but heterogeneous studies.
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34
35 *Screening*

36
37 Screening of articles will adhere to the specified inclusion criteria (as detailed above; also see Box 2).
38
39

40 **Box 2: Inclusion and exclusion criteria**

41
42
43
44 **Inclusion criteria:**

- 45 • Study population – Adults (over 18) with a diagnosis of cancer and at least one other
- 46 LTC (as specified in appendix 1), carers and health professionals
- 47 • Articles relating to experience of illness from patient, carer and professional
- 48 perspectives
- 49 • Articles focusing on areas denoted in the dimensions of interest
- 50 • Studies with a qualitative empirical design
- 51 • Articles published in English

52
53
54
55 **Exclusion criteria:**

- 56 • Articles not meeting the above inclusion criterion.
- 57 • Articles published before 2000
- 58 • Articles focusing on long term side effects of cancer treatment or second primary
- 59 cancers alone.

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10
11 A three-step screening process will be undertaken to achieve the final set of included articles. Firstly, all *titles*
12 will be read and those of no obvious relevance will be excluded. This process will be undertaken by the
13 primary reviewer DC. Secondly, *abstract screening* will be undertaken by DC and another reviewer to look in
14 more detail for relevance and fit with the inclusion criteria. Any differences in judgement will be resolved by
15 a third reviewer. Finally, *full text* articles of remaining studies will be obtained and read, again by DC and one
16 other reviewer, to assess their suitability for inclusion in the final review. Full text articles deemed to meet
17 the inclusion criteria will be selected and subject to quality appraisal and data extraction.

27 *Data extraction*

28
29
30 Data extraction will be managed in Microsoft Excel using a purpose designed proforma. Extracted material
31 will reflect the inclusion criteria and the designated aims of the review, derived from the article as a whole.
32 Information will be gathered on: author; year of publication; country of study; study type; setting; relevant
33 background and impetus for the study; methodological approach and specified methods; patient
34 characteristics and demographics including cancer and comorbidity type; main findings including pertinent
35 themes relating to experience of illness, psychosocial needs and supportive care; strengths and limitations;
36 and key relevant discussion points. Extraction of data will be carried out by DC and the second reviewer.

46 *Quality assessment*

47
48 All included articles will be subject to critical appraisal using the Critical Appraisal Skills Programme (CASP)
49 tool. The 10-item CASP tool was considered to be the most suitable tool to consider the quality parameters
50 of qualitative work and is a well validated and accepted tool. In reference to the debate on exclusion of
51 qualitative articles on the basis of quality, no articles will be excluded on these grounds alone and a low
52 threshold for inclusion will be applied so that the review can benefit from researcher insight and theoretical

1
2
3 as well as empirical contributions (28-30). The relative quality of included studies will be critically considered
4
5 during the analysis and in reference to the developed synthesis. Quality assessment will be undertaken by
6
7 two independent reviewers and any differences of opinion will be discussed and reviewed by a third person
8
9 if no consensus is reached.
10

11 12 *Data synthesis*

13
14 For a qualitative review seeking to explore personal lived experiences, it was important to find a method
15
16 that acknowledges the different paradigms and positions upon which each research output is based as well
17
18 as allowing for different study methods covering a broad and exploratory topic area (31). Meta-ethnography,
19
20 used originally in education research by Noblit and Hare but has more recently been of interest in nursing
21
22 and health services research (32-34), was considered as an appropriate method for this purpose as it is
23
24 suited to reviewing a small body of exclusively qualitative evidence and gives rise to a meaningful
25
26 interpretive account rather than simply aggregating the data (32, 35, 36). Reporting guidance for meta-
27
28 ethnography is in development (37). However, thematic synthesis, developed by Thomas and Harden,
29
30 provides a prescriptive approach that lends itself well to transparency and quality in the conduct and
31
32 reporting of the review, and in generating hypotheses to meet the review's objective of generating further
33
34 empirical research questions (21, 24). Like meta-ethnography, this method encourages the researcher to
35
36 consider the concepts and themes that relate different studies - through line by line coding, developing
37
38 descriptive themes and generating analytic themes - to provide a mutual translation and ultimately produce
39
40 an interpretive account of the phenomenon of interest, as a grounded theory approach aims to do with
41
42 primary data analysis (38, 39).
43
44
45
46
47

48 Given the anticipated yield of a small heterogeneous body of evidence meeting the eligibility criteria for the
49
50 review, a narrative-based summary may be more appropriate and achievable using steps one and two of
51
52 Thomas and Harden's thematic synthesis (21). Extracted data will be combined and described using the most
53
54 appropriate method to reflect the evidence available. Where possible, an interpretive account of the data
55
56
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1
2
3 will be presented in the context of a critical review of the evidence. The descriptive or conceptual outputs
4
5 will be related to the research questions and objectives.
6
7
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9

10 11 **DISCUSSION**

12
13 Insights from the proposed review will contribute to what is known, giving novel attention to the
14
15 combination of cancer with other long term chronic conditions. This is considered a necessary step in cancer
16
17 survivorship research and care in the face of the increasing, ageing population and the shifting emphasis in
18
19 patient care.
20

21
22 Building on the knowledge base will contribute to an evidence-based and conceptually informed integrated
23
24 model of care, with an emphasis on better understanding experiences of complex and multiple conditions
25
26 and the implications for living with cancer and comorbid illness in the push towards shared care or self-
27
28 management (40).
29
30

31
32 While conceptual and theoretical insights to a subject area can add richness to the academic evidence base,
33
34 it is important to produce a robust qualitative synthesis that reflects the identified evidence (24). A narrative
35
36 summary can adequately meet the proposed objectives, provide answers to the research questions and
37
38 speak meaningfully to policy directives.
39
40

41
42 The findings from the review will combine with patient engagement work by the authors to inform an
43
44 empirical study exploring the needs of the defined patient group and the development of an intervention to
45
46 better address their complex needs. The review will be published in a peer-reviewed journal and presented
47
48 at a relevant conference in addition to being shared with local interest groups and via online social media.
49

50
51 *Acknowledgements:* Thanks to Ms Marshall Dozier, Academic Liaison Librarian, for advice on designing the
52
53 review and searching literature databases and to Dr Karen Barnett for advice on defining included long term
54
55 conditions.
56
57
58

1
2
3 *Authors' contributions:* DC designed the systematic review with input from CC, SCB, EW and EB. DC drafted
4
5 the review protocol with input from CC, SCB, EW and EB. DC is guarantor of the review.
6
7

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9
10 reference number: PDF/15/06. The CSO have provided financial support as part of a personal fellowship but
11
12 are not involved in other aspect of the study design, implementation or analysis.
13

14
15 *Competing interests:* None declared
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Appendix 1: List of Long Term Conditions

Hypertension
Depression
Painful condition
Asthma
Coronary Heart Disease
Dyspepsia
Diabetes
Thyroid Disorders
Rheumatoid arthritis, other inflammatory polyarthropathies and systemic connective tissue disorders
Hearing loss
Chronic Obstructive Pulmonary Disease
Anxiety and other neurotic, stress-related and somatoform disorders
Irritable Bowel Syndrome
Cancer
Alcohol Problems
Other psychoactive substance misuse
Constipation
Stroke or transient ischaemic attack
Chronic kidney disease
Diverticular disease of intestine
Atrial fibrillation
Peripheral vascular disease
Heart failure
Prostate disorders
Glaucoma
Epilepsy
Dementia
Schizophrenia
Psoriasis or eczema
Inflammatory bowel disease
Migraine
Blindness and low vision
Chronic sinusitis
Learning disability
Anorexia or bulimia
Bronchiectasis
Parkinson's disease
Multiple Sclerosis
Viral hepatitis
Chronic liver disease

Source: Adapted from: Barnett K, Mercer SW, Norbury M, Watt G, Wyke S, Guthrie B. Epidemiology of multimorbidity and implications for health care, research, and medical education: a cross-sectional study. *Lancet*, 2012;380(9836):37-43.

PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) 2015 checklist: recommended items to address in a systematic review protocol*

Section and topic	Item No	Checklist item
ADMINISTRATIVE INFORMATION		
Title:		
Identification	1a	Identify the report as a protocol of a systematic review ✓ (title page, main document)
Update	1b	If the protocol is for an update of a previous systematic review, identify as such n/a
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number PROSPERO registration number: CRD42016041796 (page 5, main document)
Authors:		
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author ✓ (title page)
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review ✓ (page 11)
Amendments	4	If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments n/a
Support:		
Sources	5a	Indicate sources of financial or other support for the review ✓ (page 12)
Sponsor	5b	Provide name for the review funder and/or sponsor ✓ (page 12)
Role of sponsor or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol ✓ (page 12)
INTRODUCTION		
Rationale	6	Describe the rationale for the review in the context of what is already known ✓ (page 3, introduction)
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO) ✓ (page 4, objectives)
METHODS		
Eligibility criteria	8	Specify the study characteristics (such as PICO, study design, setting, time frame) and report characteristics (such as years considered, language, publication status) to be used as criteria for eligibility for the review ✓ (page 5)
Information sources	9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other

		grey literature sources) with planned dates of coverage ✓ (page 6)
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated ✓ (page 6-8)
Study records:		
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review ✓ (page 8)
Selection process	11b	State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (that is, screening, eligibility and inclusion in meta-analysis) ✓ (page 9)
Data collection process	11c	Describe planned method of extracting data from reports (such as piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators ✓ (page 9)
Data items	12	List and define all variables for which data will be sought (such as PICO items, funding sources), any pre-planned data assumptions and simplifications n/a
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale Dimensions of interest are listed for this qualitative synthesis rather than measurable outcomes. (page 6)
Risk of bias in individual studies	14	Describe anticipated methods for assessing risk of bias of individual studies, including whether this will be done at the outcome or study level, or both; state how this information will be used in data synthesis Quality appraisal is described (page 9)
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised n/a
	15b	If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data and methods of combining data from studies, including any planned exploration of consistency (such as I^2 , Kendall's τ) n/a
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression) n/a
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned ✓ (page 10)
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies) n/a
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE) n/a

*** It is strongly recommended that this checklist be read in conjunction with the PRISMA-P Explanation and Elaboration (cite when available) for important clarification on the items. Amendments to a review protocol should be tracked and dated. The copyright for PRISMA-P (including checklist) is held by the PRISMA-P Group and is distributed under a Creative Commons Attribution Licence 4.0.**

From: Shamseer L, Moher D, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart L, PRISMA-P Group. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. BMJ. 2015 Jan 2;349(jan02 1):g7647.

BMJ Open

Experience of living with cancer and comorbid illness: protocol for a qualitative systematic review

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Primary Subject Heading:	Oncology
Secondary Subject Heading:	Qualitative research, Health services research, Patient-centred medicine, Sociology
Keywords:	Cancer, Systematic review, Multimorbidity, Comorbidity, Qualitative synthesis

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ABSTRACT

Introduction: There is an increasing number of people living with and beyond cancer, whose experience is further complicated by additional long term health conditions in the context of an ageing population. The supportive care needs of this growing patient group should be recognised and addressed. There is a need to explore the experience of living with cancer and comorbid illness in order to develop optimal models of patient-centred care. This protocol describes a systematic review which aims to identify the qualitative evidence relating to the experience of cancer and comorbid illness for patients, informal carers and professionals, and to highlight areas where more research is needed.

Methods and Analysis: A systematic review following PRISMA guidance will be undertaken. Medline, Embase, CINAHL, PsycINFO, ASSIA, Sociological Abstracts, Web of Science, SCOPUS, OpenGrey and ProQuest Dissertations and Theses Global databases will be systematically searched for articles relevant to patient, carer and professional experiences. Two independent reviewers will screen articles for inclusion and evaluate them according to the Critical Appraisal Skills Programme (CASP) tool. Extracted data will be combined using recognised methods of qualitative synthesis to offer new insights into the topic area and for a patient-centred model of care.

Ethics and dissemination: The review does not require formal ethical review as no direct patient contact or patient identifiable data is used. Conduct of the review has been approved internally by the University of Edinburgh Centre for Population Health Sciences Ethics Review Committee. Results of the review will be published in a generalist peer-reviewed journal and presented at a relevant conference in addition to informing subsequent empirical work by the authors on this topic area.

Strengths and limitations of this study:

- Protocol for a systematic review of current literature exploring a highly important topic presenting a current challenge to health care provision
- Using robust methodology following standardised guidelines
- Scope to inform policy and practice and improve patient care
- Systematic review of qualitative empirical evidence only; does not cover quantitative studies or existing systematic reviews but can give important insights.
- Large evidence base for experience of cancer and experience of multi-morbidity, but little linking the two bodies of evidence, presenting a challenge to the reviewers.

INTRODUCTION

Due to the ageing population and better screening, diagnosis and treatments, there are an increasing number of people living with and beyond cancer (1). The lived experience of these individuals is often complicated by the presence of other comorbid chronic illnesses; as many as 78% of cancer patients are living with at least one other condition, the prevalence of which also increase with age (2, 3). Therefore, the support needs of people living with multiple complex conditions are enduring and need to be identified and addressed (4). In addition to the challenges facing secondary care (5), there is increasing recognition of the role of primary care in the provision of ongoing support to cancer patients, particularly after the completion of hospital-based treatment as patients adjust to life post treatment, living with ongoing medical issues related to their cancer treatment (6, 7). This picture becomes more complex in the presence of other long term conditions, with implications for the coordination of quality care and support (8, 9).

Meeting the needs of people living beyond cancer has been identified as a policy priority (10), which informed the development of the National Cancer Survivorship Initiative (6) in England and Wales; survivorship also forms part of the remit for Scotland's Better Cancer Care (11). In many cases survivorship initiatives and research target the clinical needs related to long term side effects (12). However, psychosocial support for survivors of cancer is another important consideration and has implications for the role of primary, secondary and community care. While valuable research has been conducted to understand better these dimensions of life for the patient, and their relatives (13-17), less is known about the impact of additional chronic illness on patients' lived experience. As such, service development and provision would benefit from further in-depth research in this area (7, 18). Understanding the challenges experienced by people living after a cancer diagnosis with other chronic conditions such as COPD, diabetes or mental ill health, can give new insights into patient-centred models of care.

Objectives

While addressing the holistic needs of cancer survivors form part of the vision of survivorship initiatives, and there is growing body of work exploring experience of multi-morbidity (19), there is a dearth of research

1
2
3 linking cancer with comorbid disease to further enrich our understanding of living with such complex illness.
4
5 This systematic review aims to synthesise and report on published evidence exploring the experience of
6
7 cancer survivorship in connection with that of other long term conditions in order to identify critical research
8
9 questions. The findings from the review will combine with those from a separate patient-centred research
10
11 prioritisation exercise to set a qualitative empirical research agenda for future work.
12
13

14 *Research questions:*

- 15
- 16
- 17 • What are the findings of qualitative evidence exploring the *experience* of living with both cancer and
- 18 one or more comorbidities from patient, carer and provider perspectives?
- 19
- 20
- 21 • What are the psychosocial support needs of people living with cancer and one or more other long
- 22 term condition(s) identified in the literature?
- 23
- 24
- 25 • What are patient, carer and provider experiences of service provision reported in the literature?
- 26
- 27
- 28 • What research priorities can be derived from the available evidence?
- 29
- 30

31 **REVIEW METHODS**

32
33
34 *Design*

35
36 A systematic review of the current evidence linking experience of cancer with comorbid illness is considered
37
38 a robust way of identifying and synthesising the published evidence in this area to define a cohesive
39
40 empirical research agenda that builds on prior knowledge (20). The review will include qualitative evidence
41
42 only to produce an interpretation built on people's views and experiences; acknowledging the rich context
43
44 and different dimensions of the lived experience from the perspective of those experiencing it. Further, a
45
46 synthesis of qualitative data aims to generate findings that are meaningful, relevant and appropriate to
47
48 individuals, to inform a qualitative research agenda, and ultimately to more effectively influence policy and
49
50 practice influencing patient outcomes (21).
51
52

53
54 The review is based on the PRISMA statement guidance for conducting a systematic review (22), and the
55
56 protocol follows the relevant items on the PRISMA-P checklist (23). The review protocol has been registered
57
58

1
2
3 on the International Prospective Register of Systematic Reviews (PROSPERO) database (registration number:
4
5 CRD42016041796).
6

7
8 The review will use methods of qualitative synthesis to combine, integrate and interpret, where possible, the
9
10 evidence from the included papers (see eligibility and data synthesis sections below)(21, 24). The review
11
12 aims to move beyond the aggregation of available data to provide further interpretive insights into living
13
14 with complex illness and define where future research can add to what is known (24).
15
16

17
18 No formal ethical approval is required for this study as it will not include patient identifiable data. It has been
19
20 internally approved by the University of Edinburgh Centre for Population Health Sciences Ethics Review
21
22 Committee (Ref: 8/03/2016).
23

24 *Eligibility criteria*

25
26 The review will include qualitative empirical studies, including unpublished grey literature. Qualitative data
27
28 from mixed methods studies will be screened for inclusion and included if the qualitative component is
29
30 relevant. Included articles will be published between 2000 and the present day to ensure the currency of the
31
32 work while enabling a broad view of developing issues to be identified. Articles will be included that address
33
34 the lived experience of the cancer journey with attention to existing long term health conditions (developed
35
36 before or after the cancer diagnosis), and will identify issues related to psychosocial and supportive care as
37
38 outlined in relation to the anticipated dimensions of interest. Articles will be included from across the cancer
39
40 continuum from diagnosis through to end of life and will include the perspectives of adult patients (aged 18
41
42 or over), informal carers and health care professionals. The review will include only articles published in
43
44 English.
45
46

47
48 Studies will be included for any cancer type in combination with one or more comorbid long term condition
49
50 (LTC), also described as cancer multi-morbidity. Long term conditions to be included will be guided by those
51
52 listed in Barnett et al's paper mapping the epidemiology of multi-morbidity (3) (See Appendix 1 for a full list
53
54 of conditions). The applied definition of long term conditions is based on ISD Scotland's report on important
55
56 long term conditions in Scotland; the report uses, among others, the Long Term Conditions Collaborative to
57
58

1
2
3 define an LTC as, 'one that requires ongoing care, limits what one can do and is likely to last longer than a
4
5 year'(25). Long term side effects of cancer treatment and second primary cancers are not included;
6
7 experience of second primary cancers is being addressed separately within the research centre. Where it is
8
9 not possible to determine whether the condition is caused by cancer treatment, the article will be included
10
11 in the review but analysed separately.
12

13 *Dimensions of interest*

14
15 The review focuses on an exploration of the dimensions of lived experience: physical, social, emotional and
16
17 psychological. From a similar study exploring multiple dimensions of experience, these are expected to
18
19 include such topics as quality of life, well-being, psychosocial supportive care needs, attitude and identity,
20
21 perceived continuity and quality of care, and influence of personal demographics (age, gender, ethnicity,
22
23 socio-economic status) (26). In relation to how services can best meet patients' needs, topics may include
24
25 issues of accessing appropriate services, coordination of care for multiple conditions, and self-management
26
27 across the cancer care continuum (17, 19) . The search strategy has been developed to reflect these broad
28
29 and exploratory domains. The outcomes will depend on the evidence available and gaps in the evidence will
30
31 be highlighted for future study.
32
33
34

35 *Information sources*

36
37 A variety of search strategies will be used to identify potential literature for the review, particularly given
38
39 the challenges in identifying qualitative evidence (27). This will primarily involve a literature database search
40
41 (including grey literature), but will also include citation and snowball searching, known expert consultation
42
43 via email, related articles searches in PubMed and use of Google scholar. The databases to be consulted are:
44
45 Medline, Embase, CINAHL, PsycINFO, ASSIA, Sociological Abstracts, Web of Science, SCOPUS and, for grey
46
47 literature, OpenGrey and ProQuest Dissertations and Theses Global. These are considered sufficient to
48
49 comprehensively cover the range of topics and disciplines implicated in this review.
50
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Search strategy

The searching phase aims to identify relevant studies for inclusion in the review. The process of searching will begin with individually searching each of the listed databases using an adapted list of search terms; the developed search strategy for Medline is shown in Box 1. The search terms will be adapted to suit each database to derive the most meaningful search and will use free text, MeSH and subject headings for maximum sensitivity and specificity.

Box 1: Example search strategy for Medline

1. Comorbidity/
2. (Multimorbid\$ or multi-morbid\$ or Co-morbid\$).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
3. Chronic Disease/ or (Chronic adj5 (illness\$ or condition\$ or disease)).tw.
4. (Long term adj5 (condition\$ or illness\$ or disease\$)).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
5. 1 or 2 or 3 or 4
6. Neoplasms/ or Cancer.mp or (Tumour or tumor or Oncology or Neoplasm).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
7. Qualitative Research/ or Qualitative.mp or Interview/ r Interview\$.mp or In-depth.mp
8. (Focus Group or or Ethnograph\$ or Observation\$ or Participant\$ or Respondent\$ or View\$ or Belief\$ or Attitude\$ or Awareness or Perspective\$ or Understanding\$ or Findings or Grounded Theory or Social Construction\$ or Theoretical or Phenomenolog\$).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
9. 7 or 8
10. Psychosocial.mp. or Stress, Psychological/ or Social Support/ or Psycho-social.mp or "Quality of Life"/ or "Experience of Illness".mp or Experience adj5 Illness or Attitude to Health/ or "Quality of Life".mp or Happ\$.mp or Emotion\$.mp. or Emotions/
11. Patient satisfaction.mp. or Patient Satisfaction/ or "Quality of Health Care"/ or "Patient Experience".mp or Satisfaction.mp or "Information Preferences".mp or Decision Making/ or (Decision-making or Decision Making).mp or (Informed choice or Informed Decision Making).mp or Self management.mp. or Self Care/ or "Continuity of Patient Care"/ or Integrated care.mp
12. Access to Health Care.mp. or Health Services Accessibility/
13. Identity.mp. or gender.mp. or Gender Identity/
14. (Health behaviour or Health behavior).mp.
15. (Help-seeking or Help seeking).mp.
16. (Well-being or Well being).mp. or Holistic.mp or Continuity of Care.mp or "Continuity of Patient Care"/ or (Fragmented adj5 care).mp or (joined-up or Joined Up).mp or Integrated care.mp or Diversity of Care.mp.
17. Primary care.mp. or Primary Health Care/ or Secondary care.mp. or Secondary Care/ or Community Care.mp or Community Health Services/ or Ambulatory Care.mp. or Ambulatory Care/ or "Delivery of Health Care, Integrated"/ or Preventive Health Services/ or Family Practice/ or Shared care.mp or Collaborat\$ care.mp

1
2
3
4 18. (Patient-centred care or Patient centred care or Patient-centred or Person-centred or Patient centred or Person
5 centred).mp or Patient-Centered Care/

6 19. Palliative Care/ or Supportive care.mp. or palliative.mp.

7
8 20. "Health Services Needs and Demand"/ or unmet need.mp. or Health Services Research/ or Depriv\$.mp. or (Socio-
9 economic or Socioeconomic).mp. or Information needs.mp.

10 21. Survivor\$.mp. or Survivors/

11 22. Patient Care Planning/ or Care plan.mp. or (Follow-up or Follow up).mp.

12 23. ("use of service\$" or "service use" or "service adj5 use").mp.

13 24. 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23

14 25. 5 AND 6 AND 9 AND 24
15
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18
19
20 **DATA COLLECTION AND ANALYSIS**

21 *Study records*

22 Identified records will be imported into and managed in EndNoteX7. Screened and selected articles will be
23 managed in subsequent EndNote databases in order to track and record the number of records retained at
24 each step. It is anticipated that given the relative lack of attention in the literature specifically examining the
25 experience of long term conditions as a cancer survivor, the review will produce a small number of relevant
26 but heterogeneous studies.
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35 *Screening*

36 Screening of articles will adhere to the specified inclusion criteria (as detailed above; also see Box 2).
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40 **Box 2: Inclusion and exclusion criteria**

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44 **Inclusion criteria:**

- 45 • Study population – Adults (over 18) with a diagnosis of cancer and at least one other
- 46 LTC (as specified in appendix 1), carers and health professionals
- 47 • Articles relating to experience of illness from patient, carer and professional
- 48 perspectives
- 49 • Articles focusing on areas denoted in the dimensions of interest
- 50 • Studies with a qualitative empirical design
- 51 • Articles published in English

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55 **Exclusion criteria:**

- 56 • Articles not meeting the above inclusion criterion.
- 57 • Articles published before 2000
- 58 • Articles focusing on long term side effects of cancer treatment or second primary
- 59 cancers alone.

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11 A three-step screening process will be undertaken to achieve the final set of included articles. Firstly, all *titles*
12 will be read and those of no obvious relevance will be excluded. This process will be undertaken by the
13 primary reviewer DC. Secondly, *abstract screening* will be undertaken by DC and another reviewer to look in
14 more detail for relevance and fit with the inclusion criteria. Any differences in judgement will be resolved by
15 a third reviewer. Finally, *full text* articles of remaining studies will be obtained and read, again by DC and one
16 other reviewer, to assess their suitability for inclusion in the final review. Full text articles deemed to meet
17 the inclusion criteria will be selected and subject to quality appraisal and data extraction.
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27 *Data extraction*

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29 Data extraction will be managed in Microsoft Excel using a purpose designed proforma. Extracted material
30 will reflect the inclusion criteria and the designated aims of the review, derived from the article as a whole.
31 Information will be gathered on: author; year of publication; country of study; study type; setting; relevant
32 background and impetus for the study; methodological approach and specified methods; patient
33 characteristics and demographics including cancer and comorbidity type; main findings including pertinent
34 themes relating to experience of illness, psychosocial needs and supportive care; strengths and limitations;
35 and key relevant discussion points. Extraction of data will be carried out by DC and the second reviewer.
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45 *Quality assessment*

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47 All included articles will be subject to critical appraisal using the Critical Appraisal Skills Programme (CASP)
48 tool. The 10-item CASP tool was considered to be the most suitable tool to consider the quality parameters
49 of qualitative work and is a well validated and accepted tool. In reference to the debate on exclusion of
50 qualitative articles on the basis of quality, no articles will be excluded on these grounds alone and a low
51 threshold for inclusion will be applied so that the review can benefit from researcher insight and theoretical
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3 as well as empirical contributions (28-30). The relative quality of included studies will be critically considered
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5 during the analysis and in reference to the developed synthesis. Quality assessment will be undertaken by
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7 two independent reviewers and any differences of opinion will be discussed and reviewed by a third person
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9 if no consensus is reached.
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11 12 *Data synthesis*

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14 For a qualitative review seeking to explore personal lived experiences, it was important to find a method
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16 that acknowledges the different paradigms and positions upon which each research output is based as well
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18 as allowing for different study methods covering a broad and exploratory topic area (31). Meta-ethnography,
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20 used originally in education research by Noblit and Hare but has more recently been of interest in nursing
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22 and health services research (32-34), was considered as an appropriate method for this purpose as it is
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24 suited to reviewing a small body of exclusively qualitative evidence and gives rise to a meaningful
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26 interpretive account rather than simply aggregating the data (32, 35, 36). However, thematic synthesis,
27
28 developed by Thomas and Harden, was also considered as it provides a prescriptive approach that lends
29
30 itself well to transparency and quality in the conduct and reporting of the review, and in generating
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32 hypotheses to meet the review's objective of generating further empirical research questions (21, 24). Like
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34 meta-ethnography, this method encourages the researcher to consider the concepts and themes that relate
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36 different studies - through line by line coding, developing descriptive themes and generating analytic themes
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38 - to provide a mutual translation and ultimately produce an interpretive account of the phenomenon of
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40 interest, as a grounded theory approach aims to do with primary data analysis (37, 38).
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46 Given the anticipated yield of a small heterogeneous body of evidence meeting the eligibility criteria for the
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48 review, a narrative-based summary may be more appropriate and achievable using steps one and two of
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50 Thomas and Harden's thematic synthesis (21). Extracted data will be combined and described using the most
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52 appropriate method to reflect the evidence available. Where possible, an interpretive account of the data
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54 will be presented in the context of a critical review of the evidence. The descriptive or conceptual outputs
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56 will be related to the research questions and objectives.
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DISCUSSION

Insights from the proposed review will contribute to what is known, giving novel attention to the combination of cancer with other long term chronic conditions. This is considered a necessary step in cancer survivorship research and care in the face of the increasing, ageing population and the shifting emphasis in patient care.

Building on the knowledge base will contribute to an evidence-based and conceptually informed integrated model of care, with an emphasis on better understanding experiences of complex and multiple conditions and the implications for living with cancer and comorbid illness in the push towards shared care or self-management (39).

While conceptual and theoretical insights to a subject area can add richness to the academic evidence base, it is important to produce a robust qualitative synthesis that reflects the identified evidence (24). A narrative summary can adequately meet the proposed objectives, provide answers to the research questions and speak meaningfully to policy directives.

The findings from the review will combine with patient engagement work by the authors to inform an empirical study exploring the needs of the defined patient group and the development of an intervention to better address their complex needs. The review will be published in a peer-reviewed journal and presented at a relevant conference in addition to being shared with local interest groups and via online social media.

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Authors' contributions: DC designed the systematic review with input from CC, SCB, EW and EB. DC drafted the review protocol with input from CC, SCB, EW and EB. DC is guarantor of the review.

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Competing interests: None declared

For peer review only

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Appendix 1: List of Long Term Conditions

Hypertension
Depression
Painful condition
Asthma
Coronary Heart Disease
Dyspepsia
Diabetes
Thyroid Disorders
Rheumatoid arthritis, other inflammatory polyarthropathies and systemic connective tissue disorders
Hearing loss
Chronic Obstructive Pulmonary Disease
Anxiety and other neurotic, stress-related and somatoform disorders
Irritable Bowel Syndrome
Cancer
Alcohol Problems
Other psychoactive substance misuse
Constipation
Stroke or transient ischaemic attack
Chronic kidney disease
Diverticular disease of intestine
Atrial fibrillation
Peripheral vascular disease
Heart failure
Prostate disorders
Glaucoma
Epilepsy
Dementia
Schizophrenia
Psoriasis or eczema
Inflammatory bowel disease
Migraine
Blindness and low vision
Chronic sinusitis
Learning disability
Anorexia or bulimia
Bronchiectasis
Parkinson's disease
Multiple Sclerosis
Viral hepatitis
Chronic liver disease

Source: Adapted from: Barnett K, Mercer SW, Norbury M, Watt G, Wyke S, Guthrie B. Epidemiology of multimorbidity and implications for health care, research, and medical education: a cross-sectional study. *Lancet*, 2012;380(9836):37-43.

PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) 2015 checklist: recommended items to address in a systematic review protocol*

Section and topic	Item No	Checklist item
ADMINISTRATIVE INFORMATION		
Title:		
Identification	1a	Identify the report as a protocol of a systematic review ✓ (title page, main document)
Update	1b	If the protocol is for an update of a previous systematic review, identify as such n/a
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number PROSPERO registration number: CRD42016041796 (page 5, main document)
Authors:		
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author ✓ (title page)
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review ✓ (page 11)
Amendments	4	If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments n/a
Support:		
Sources	5a	Indicate sources of financial or other support for the review ✓ (page 12)
Sponsor	5b	Provide name for the review funder and/or sponsor ✓ (page 12)
Role of sponsor or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol ✓ (page 12)
INTRODUCTION		
Rationale	6	Describe the rationale for the review in the context of what is already known ✓ (page 3, introduction)
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO) ✓ (page 4, objectives)
METHODS		
Eligibility criteria	8	Specify the study characteristics (such as PICO, study design, setting, time frame) and report characteristics (such as years considered, language, publication status) to be used as criteria for eligibility for the review ✓ (page 5)
Information sources	9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other

		grey literature sources) with planned dates of coverage ✓ (page 6)
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated ✓ (page 6-8)
Study records:		
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review ✓ (page 8)
Selection process	11b	State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (that is, screening, eligibility and inclusion in meta-analysis) ✓ (page 9)
Data collection process	11c	Describe planned method of extracting data from reports (such as piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators ✓ (page 9)
Data items	12	List and define all variables for which data will be sought (such as PICO items, funding sources), any pre-planned data assumptions and simplifications n/a
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale Dimensions of interest are listed for this qualitative synthesis rather than measurable outcomes. (page 6)
Risk of bias in individual studies	14	Describe anticipated methods for assessing risk of bias of individual studies, including whether this will be done at the outcome or study level, or both; state how this information will be used in data synthesis Quality appraisal is described (page 9)
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised n/a
	15b	If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data and methods of combining data from studies, including any planned exploration of consistency (such as I^2 , Kendall's τ) n/a
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression) n/a
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned ✓ (page 10)
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies) n/a
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE) n/a

*** It is strongly recommended that this checklist be read in conjunction with the PRISMA-P Explanation and Elaboration (cite when available) for important clarification on the items. Amendments to a review protocol should be tracked and dated. The copyright for PRISMA-P (including checklist) is held by the PRISMA-P Group and is distributed under a Creative Commons Attribution Licence 4.0.**

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