

EXPERTS II - How are patient and caregiver participation in health and social care shaped by experienced burden of treatment and social inequalities? Protocol for a qualitative synthesis (NIHR HS&DR 130407)

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1. ABSTRACTS

1.1 SCIENTIFIC ABSTRACT

Research question: how are service user and caregiver participation in health and social care shaped by experienced burden of treatment and social inequalities?

Background: The *workload* service users and caregivers take on, and their *capacity* to do this work, when they engage with and participate in different kinds of care is important. It may play a key part in shaping the adoption and implementation of innovations in service delivery and organisation, utilisation and satisfaction with services, and the outcomes of care. This is reflected in policy and practice interventions that identify service users and caregivers as part of a *team* that consists of informal networks beyond provider organisations and the professionals within them in health and social care. Much work in this field has been aimed at service user and caregiver experiences of a narrow range of long-term conditions, and these studies have often focused on factors that motivate and shape adherence to self-care regimes.

Aims and objectives: To review, compare, and synthesise qualitative studies of the lived experience of the *work* of service user and caregiver engagement and participation in mixed economies of self-care, health and social care that are differently shaped by interactions between experienced treatment burdens and social inequalities. To derive policy and practice implications for service redesign. Including: qualitative studies of the lived experience of three kinds of condition: long-term conditions associated with significant disability (Parkinson's disease, schizophrenia); serious relapsing remitting disease (ulcerative colitis, bipolar disorder); and rapidly progressing acute disease (brain cancer, rapidly progressing dementia).

Methods: Qualitative evidence synthesis. The review will be conducted in four phases:

- Construction of a theory-informed coding manual to support comparative analysis of documentary data; systematic search of bibliographic databases to identify, screen and collect full-text papers;
- Analysis of texts of relevant studies using conventional manual coding techniques, and text mining software; construction of taxonomies of service user and caregiver work and capacity;
- Model core components and identify common factors across index conditions, disease trajectories, and service contexts.
- Working with a reference group of health and social care professionals, and a PPI group, to explore the validity of the models produced by the review, to develop workload reduction strategies, and to consider person-centered service design for people with complex health problems.

Timelines for delivery: Coding manual drafted, searches completed, and screening begun by month six. Manual coding and text mining commences in month four and will be completed by month 20. Preliminary reporting and dissemination commences at month 18. Final report completed and submitted at Month 24.

Anticipated impact and dissemination: Practitioners and researchers will be better placed to understand structural factors that shape treatment burden and affect service user activation. The second impact of this research will be to identify promising targets for service design, and for policy restructuring. Dissemination strategies include the promotion of workload reduction models to support service users and caregivers and their advocates, and policy briefs and peer-reviewed reports for practitioners, policy makers, and researchers.

1.2 PLAIN ENGLISH SUMMARY OF RESEARCH

Our experiences of illness are often complex. We may have to work hard too. This work can take the form of tasks that are delegated to us by health and social care providers. When we meet health and care professionals we often find ourselves agreeing with them about the kinds of work that we need to do. These are called *negotiated obligations*. We may need to monitor, manage and record symptoms; take up different diets and physical activity; obtain and use different drugs, dressings and medical devices; develop expertise in using websites and information technology; seek help, access and coordinate input and support from health and social care services; sometimes we have to work out how we are going to pay for the services that we need. How we get through the work is affected by our capacity to do it, and that is shaped by the different personal and wider resources that we can draw on. The work that we do does not just involve our negotiated obligations with health and social care professionals and organisations. It goes beyond that to include the things that we and our caregivers, family, friends and others volunteer to do to make our healthcare possible. These are called *assumed obligations*. All of this is also affected by the services that are available to us, and by the ways in which our chances in life are shaped by our income, ethnicity, education, gender, and age. The kinds of illnesses we have, how they progress, and the demands that these make on us, mean that these factors change over time. We call these changes *trajectories*. Most research on service user work has looked at a group of chronic diseases with long trajectories and gradually declining service user capacity. This means that much less is known about how service user and caregiver work plays out in other complex trajectories, how they are shaped by different kinds of services, and how people are impacted by different kinds of social and economic disadvantage.

To better understand service user work and capacity, and the different forces that shape them, we propose to perform an ambitious review of published studies that have sought to tell us about people's everyday experiences of living with illnesses with three kinds of trajectory. These are long-term conditions associated with significant disability; serious relapsing remitting disease; and rapidly progressing acute disease. We will start by drawing on existing research to build a framework in which we can identify, describe, and understand relevant aspects of published studies. Then we will extract relevant information from them using two different techniques. We will do this by carefully reading and manually identifying and extracting key data, and we will also use a computer program that looks for particular kinds of words and phrases and finds links between them. This is called text mining. Combining manual analysis and text mining will enable us to make a comprehensive map or model of common features of service user work and capacity across different kinds of conditions, their trajectories, service organisation and delivery, and patterns of social and economic disadvantage. Finally, we will work with groups of service users, caregivers and their advocates, and with health and social care professionals and managers to apply the model to the development of strategies to reduce workload and promote service redesign for people with complex health problems.

2. INTRODUCTION

The *workload* that service users[†] and caregivers take on, and their *capacity* to do this work, when they engage with and participate in different kinds of care is important and may play a key part in shaping the adoption and implementation of innovations in service delivery and organisation, utilisation and satisfaction with services, and the outcomes of care.¹ This is reflected in policy and practice interventions that identify service users and caregivers as part of a *team* that consists of informal networks beyond provider organisations in health and social care health and care, as well as professionals within them.² Much work in this field has been aimed at service user and caregiver experiences of a narrow range of complex long-term conditions, and these studies have often focused on adherence to self-care regimes.³ In this review we will extend our understanding of service user and caregiver workload and capacity by comparing the ways that they are revealed in qualitative studies of the lived experience of a wider variety of physical and mental health problems characterised by long-term, relapsing remitting, and rapidly progressing trajectories. Using both conventional models of qualitative analysis and novel text mining approaches, we will explore the ways in which these experiences are shaped by interactions with self-care, healthcare, and social care professionals and provider organisation; by patterns of service organisation and delivery; and by different kinds of inequalities of access and provision of care, long with wider social inequalities. This evidence synthesis will create a taxonomy of service user and caregiver work associated with lived experiences of burden of treatment; a taxonomy of theoretical constructs that explain interactions between them; and identification of core components of service user and caregiver experience of configurations of care. These will support development and implementation of new, service user-centred models of care.

3. BACKGROUND AND RATIONALE

3.1 Service users and caregivers do work when they engage with self-care, healthcare, and social care. Over five decades, empirical research in medical sociology and social psychology, health psychology, and medical anthropology has led to a very large body of literature that points to the importance of service user and caregiver *work* in shaping engagement, participation, survivorship, and clinical and social outcomes across a range of healthcare problems.¹ This corpus of studies has drawn attention to work that is intimately linked to *being* (negotiating experience and identity)⁴; *changing* (managing status passage and biographical disruption)^{5,6}; *relating* (participating in interactions within healthcare provision and informal social networks)⁷; and *doing* (performing health behaviours and enacting healthcare technologies and self-care practices)⁸. The work that service users and caregivers do has always been important, but is becoming more so because healthcare provider organisations are increasingly promoting models of care in which service users and caregivers are seen to be integral to the healthcare team and thus the workforce.

3.2 What do we mean by service user work? This review focuses on service user and caregiver work and capacity, across a range of complex disease types and trajectories. Its main focus is on burden of treatment and the ways that it shapes the experience of service users and caregivers.

[†] **Note added on 18 November 2020.** In our application for funding, we used the term ‘patient’ throughout to define participants in health and social care processes. For reasons outlined in Section 3.2.1 throughout this protocol we use the term ‘service user’ to define participants. The project title remains unchanged to ensure consistency with NIHR documentation and records.

3.2.1. Definition of participants. Any study that investigates the experiences of people who use multiple services faces the problem of acknowledging and supporting the complex ways that they self-identify. People who are involved in self-care, or who are in remission from relapsing remitting diseases, may not see themselves as service users. People using formal health services may be called service users, but people who use mental health services, or who have recovered from other serious illnesses, may call themselves survivors. People who identify with the disability movement may want to use terminology that emphasises control. People using social care services may be called customers, clients or service users, but may self-identify in very different ways. In this proposal, but not in the review itself, we use the term *service user*. We use service user to mean someone who is sick, who is in a relationship with one or more health or social care services because of that sickness, and whose experience of that service is shaped by social inequalities of some kind. The term caregivers may refer to partners and spouses, other family members, friends, employees of service provider organisations, and even holders of power of attorney, official guardians and other officers of the court. The important feature of a caregiver is that they perform affective, cognitive, informational and material work with and for service users.

3.2.2 Definition of service user and caregiver workload. Service users and caregivers take on multiple tasks when they participate in care. These tasks include those that arise from negotiated obligations to participate in delegated clinical work, such as conforming to expectations of behaviour modification and change; symptom monitoring and management; adhering to complex treatment regimens and managing multiple drugs, dressings, medical devices, web-enabled tools and information sources, and prostheses. Beyond these negotiated obligations to perform clinical work that is handed off to service users and caregivers, are the *assumed obligations* that arise when service users and caregivers have to take on the organisational work that they need to do to engage, and stay engaged, with health and social care providers. This includes tasks relating to participation, accessing, navigating, coordinating and managing processes of care with (often uncoordinated) multiple service providers and their complex administrative systems and care pathways.⁹⁻¹¹

3.2.3 Definition of service user and caregiver capacity. Service users and caregivers have finite capacity. We define capacity as the combination of affective, cognitive, relational, informational, material and economic resources available to the service user that make it possible for them to participate in care and to meet the normative expectations of provider organisations and professionals.¹¹ Capacity is not just a function of individual characteristics of service users and caregivers, but is shaped by the *social and relational contexts* in which they are located. They experience structural social advantage and disadvantage; varying access to supportive social networks and social capital¹²; and in their immediate social relations they draw on the collective resilience,¹³ competence,¹ and efficacy¹⁴ of network members. Capacity is also shaped by the pathophysiological, psychological, cognitive and emotional effects of disease as these play out over time¹⁵; by social and institutional responses to particular forms of ill-health that include changes in social status, stigma, and assessments of culpability and the legitimacy of different expressions of symptoms.

3.2.4 Definition of trajectories. Health and social care services increasingly characterise the ways that people move through them and access care using the language of processes and pathways. Clinical pathways are system-level tools for organising service users according to diagnosis, treatment modality, professional contact, and disease progression. Pathways differ between different specialisms and healthcare provider organisations.¹⁶ However, they may not be configured or understood in the same way by social care providers. For service users and caregivers,

the situation may be more complex.¹⁷ Disease progression itself may constitute a temporal trajectory. In some diseases, trajectories may run over many years and be associated with cumulative and often significant disability, as in Parkinson's disease. In relapsing-remitting diseases like bipolar disorder trajectories may oscillate between recovery and recurrence and the future is characterised by uncertainty. In rapidly progressive acute diseases, such as some brain cancers, trajectories involve significant and often acute pathophysiological deterioration characterised by rapid loss of physical and mental capacity and struggles over rescue and recovery. These trajectories are more than changes that take place over time, and they are often more than the sum of pathophysiological processes. Instead, they may take the form of status passages,⁵ in which service users and caregivers' social identities are formed and changed according to the ways in which others relate to the character and effects of their illness, the degree of disruption to relationships and socio-economic status that follow from it, and anticipated outcomes of disease progression.⁶

3.3 Sources of fragmentation in service provision. Interest in the conduct of service user and caregiver work reflects the ways that its character has changed as healthcare providers around the world have had to respond to an epidemiological transition from acute and infectious disease, to long-term, non-communicable, and comorbid conditions.¹⁸ These conditions are often associated with ageing. They are exacerbated by social inequalities, and there is good evidence that economic and educational inequalities are associated with earlier onset of comorbid long-term conditions.¹⁹ In response to these problems, innovations in the organisation and delivery of care have increasingly shifted locus of clinical activity away from individualised interactions between doctors and service users characterised by continuity of care, to transactional models of care in which service users encounter multiple service providers who perform specific technical tasks and who are located in complex technical divisions of labour.²⁰ It is not clear whether the same situation applies in social care, but as service commissioners and providers have come under pressure to reduce costs and outsource services considerable fragmentation in service organisation and delivery has become evident. Service users and caregivers may thus be located in mixed economies of care provision that includes in NHS, statutory social care, private sector, and third sector providers.

3.4 Structural disadvantage affects participation. Complexity in experiences of health and social care, and of health and social care systems needs also to be understood in the context of well-established effects of socio-economic disadvantage, along with other structural disadvantages formed around gender, ethnicity, age, and migration. The role of structural inequalities in forming a context for sometimes hard and heavy work for service users and caregivers is well established.^{21,22} Groups exhibiting structural advantages experience better health; fewer comorbidities; and later onset of chronic comorbidities. Importantly, observed interactions between advantaged populations and health services are characterised by experiences of better quality and easier access to healthcare, better access to formal and informal support mechanisms, and fewer environmental stressors.²³ The health and social care landscape is shaped by unevenly distributed structural advantage, system-level forces, and epidemiological changes. These profoundly affect expectations of service user and caregiver contributions to their care, indeed they may experience extremes of disadvantage.²⁴

3.5 Expectations of service user and caregivers. Changes in the character of service user and caregiver work in healthcare call for a radical reconsideration of their roles: indeed, policy and research initiatives link engagement in these processes of care to explicit expectations of participation in a healthcare 'team' or informal membership of the healthcare 'workforce'.² These expectations of participation call for very different investments by service users and caregivers, in which individual motivation and adherence to treatment regimens are likely profoundly influenced by the workload that stem from them and their capacity to meet its demands.¹⁵ These investments are sometimes contested by service users and caregivers.²⁵

These can require significant numeracy and literacy, as well as high level administrative and technological skills, in vulnerable and disadvantaged populations.²⁶ This work is often distributed within complex social networks and relational processes.¹⁸ It thus calls for collective action, efficacy, and competence over and above individual psychological variables such as self-efficacy.^{25,27-31} It is not clear how these expectations and interactions correspond to those of social care, but it is likely that these are also unevenly distributed, and this unevenness may give rise to important inequalities in capacity, participation, and outcomes.³²

3.6 Work leading up to this proposal.

Following on from the germinal research noted in section 3.1, we have contributed to this literature. We have pointed to the importance of the *workload* that service users and caregivers take on when they have to manage their health and healthcare,³³ and when they have to understand and organise their interactions with

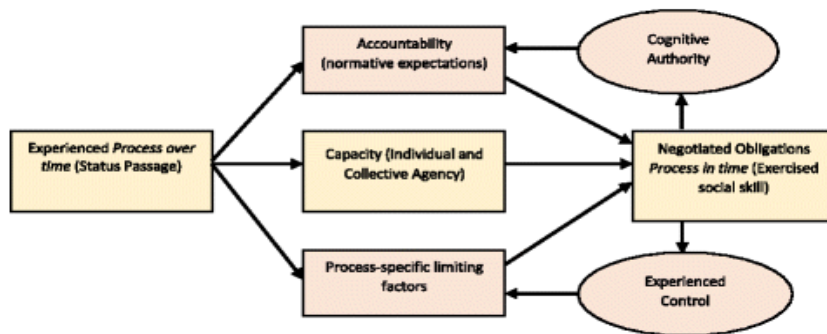


Figure 1: Accountability, capacity and negotiated obligations

healthcare agencies and other entities.³⁴ We have explored how self-care and healthcare workload can burden service users and their families, and how capacity to handle this workload varies between individuals. Our contributions to this have been through theoretical development as well as empirical research. Building normalization process theory has helped us understand the ways in which experiences of service user-hood can be understood as material and relational work that moves back and forth between the clinic and home; the cumulative complexity model, has helped us to understand service user workload and capacity over linear time and proposed that is associated with poor healthcare utilisation and outcomes; and burden of treatment theory has helped us to understand the distribution of service user and caregiver workload and capacity over relational and organisational space.¹ These developments enable us to model burden of treatment as a result of micro-level phenomena in which material and interactional practices are allocated and negotiated in complex interactions between people, disease processes, and healthcare environments.^{10,32,35-38} (See **figure 1**.¹¹) Understanding service user capacity, balancing preferences, and controlling workload allocation and capacity will support the design of minimally disruptive models of care that work across sectors.

4. WHY IS THIS RESEARCH NEEDED NOW?

4.1 Why do we need another systematic literature review? Much is now known about experiences of treatment burden in specific long-term conditions (especially diabetes, heart failure, COPD and chronic kidney disease, stroke) because they are common diseases that generate high levels of demand, consequent workload and expenditure, and are targets for self-care interventions intended to promote service user activation and slow down disease progression. This had made them important foci for research. But focusing on conditions characterised by trajectories of disease progression and degeneration over several years means that important features of other kinds of illness are less visible. It is clear, for example,

that experiences of symptoms and care, workload and capacity, are very different in disease of long duration (e.g. COPD) and relatively rapid progression (e.g. lung cancer) although these diseases have similar effects and are equally lethal.³⁹ Much less is known about the ways that workload and capacity are constituted and experienced in mental health problems.

4.2 Is a review of multiple disease types and trajectories, service contexts, and social contexts feasible?

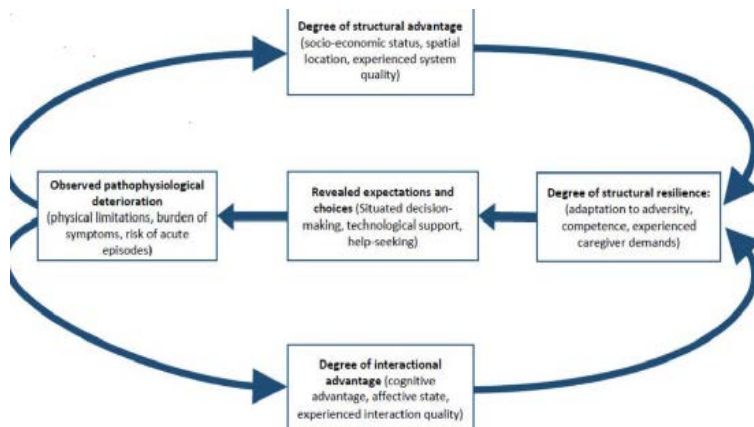


Figure 2: EXPERTS I Model of Workload/Capacity interactions during pathophysiological deterioration

To build more person-centred and responsive services we need to understand interactions between (a) service user and caregiver experiences; (b) service organisation and delivery; and (c) structural and system-level patterning of advantage. Although complex comparative qualitative syntheses are challenging to perform, we have previously demonstrated that such an approach is feasible and rewarding. In the EXPERTS I

review^{40 41} we analysed and compared reviews of qualitative studies of lived experience of heart failure, COPD, and chronic kidney disease. This showed that the factors we wish to address are important. However, that review focused on the impact of factors related to management of workload and capacity in conditions marked by significant pathophysiological deterioration towards the end of life. It produced a preliminary model (see **figure 2 above**⁴¹), that can form the starting point for our proposed review.

4.3 The need for comparative analyses. Our proposed systematic review of qualitative studies of experiences of self-care, health and social care is an important step towards modelling interactions between services and their contexts, developing instrumentation, and developing and evaluating interventions at the individual and organisational levels that will support service users and caregivers, and that will support demand management strategies at a system level. The review will focus on disease types and trajectories rather than single index conditions: we are interested in the ways that work and capacity are played out differently across a space characterised by different service providers and different patterns of social inequality, rather than by a specific clinical problem.

4.3.1 Understanding service user experience beyond single index conditions. Our comparator conditions are physical and mental health problems that are defined by one of three trajectories. These are: *long-term conditions associated with significant disability, relapsing remitting disease, and rapidly progressive acute disease.*

4.3.2 Parity between condition types. The review gives parity to physical and mental health problems, while recognising that these are experienced in different ways. For example, experienced workload and capacity are likely to be very different in schizophrenia and astrocytoma, in part because of differences in service organisation and delivery and social context.

4.3.3 Parity between condition trajectories. Most work on service user work, workload, and capacity has focused on conditions of relatively long duration in which behaviour modification and self-care are important components of management. Much less is known about relapsing-remitting conditions and the different workload and

capacity problems that stem from them, or about diseases that progress rapidly to conclusion, in which workload and capacity may be transferred from service users to caregivers quite early in their trajectory.

4.3.4: Multiple focal points. Comparative analyses will reveal a core set of constructs which vary between index conditions, service contexts, and disease trajectories. They will also reveal the important differences in the ways in which workload and capacity are experienced across index conditions, three disease trajectories, and three service contexts.

5. AIMS AND OBJECTIVES

5.1 AIM

We will review, compare, and synthesise qualitative studies of the lived experience of physical and mental health problems characterised by long-term, relapsing remitting, and rapidly progressing trajectories. In these contexts, we will (a) investigate the work of service user and caregiver engagement and participation in self-care, health and social care; (b) understand how these are differently shaped by interactions between burden of treatment and social inequalities, and (c) provide a platform for responsive service design.

5.2 OBJECTIVES

5.2.1 Identification of studies. We will systematically review and search for qualitative studies of the lived experience of three kinds of condition: *long-term conditions associated with significant disability* (Parkinson's disease and schizophrenia); *serious relapsing remitting disease* (ulcerative colitis, bipolar disorder); *rapidly progressing acute disease* (brain cancer, rapidly progressing dementia).

5.2.2 Qualitative analysis. Within materials included in the review, we will; (a) identify the work of service user and caregiver engagement and participation in self-care, health and social care; (b) characterise how these are differently shaped by interactions between burden of treatment (negotiated and assumed obligations), and social inequalities, and (c) understand the elements of these that could contribute to responsive service design.

5.2.3 Theoretical development. We will develop from this literature review (a) a taxonomy of service user and caregiver work associated with lived experiences of different condition types and trajectories, (b) a taxonomy of theoretical constructs that explain interactions between condition types and trajectories, service contexts and social inequalities and (c) a translational framework to support the development and implementation of new, person centred models of care for service users and caregivers.

6. RESEARCH PLAN[‡]

[‡] **Note added on 18 November 2020.** The version of this protocol that was peer-reviewed and approved for funding preceded the COVID-19 epidemic. However, the epidemic has affected important preliminary aspects of the work. The approved protocol included a series of four workshops with health and social care professionals and researchers (in June and October 2020, and in March and November 2021) to assist in the development of the coding strategy and to explore the insights that developed from this. The COVID-19 epidemic made in-person

6.1 Design and theoretical/conceptual framework. Following the procedures we developed for the EXPERTS I study^{40 41} we will perform a theory-informed synthesis of qualitative studies. The study will take 24 months, and employ conventional ‘manual’ qualitative analysis.⁴² We will add a new dimension to our work by using Leximancer® software to perform text mining across the data set. In both text mining and manual analysis, we will explore the extent to which constructs are present across the qualitative data set, or whether they are concentrated around particular index conditions or disease trajectories.

6.1.1 Linking healthcare constructs with social care literature. An important theoretical and methodological problem in this review is synthesising research literature from different fields in which different technical vocabularies and theoretical constructs are employed. We expect to find differences in the ways that health and social care researchers identify, characterise and explain key constructs. We need to understand this better as we produce a coding manual. As our coding frame is developed, we will seek advice from professionals and service users across the health and social care field. Beginning with our oversight group, we will use a variety of techniques to identify useful discussants, and we will also use social media channels to identify experts by experience and academic experts to contribute to this process.

6.1.2 Development of a coding manual for the review. Because we will be using Leximancer software that performs semantic and relational searches, we need to create at the outset a coding manual or lexicon for the whole study. This will involve developing a set of theory-informed terms (e.g. Strauss et al’s concept of articulation work,⁸ or Vassilev et al’s concept of collective efficacy¹⁴) that can be translated into everyday language and then used to search text. To do this, we will draw on three bodies of theory:

- (i) Core concepts in the writings of Anselm Strauss and colleagues in the US that has set out interactionist models of ‘work’ as an integral element of the lived experience of health and illness,^{8,43,44} and status passage theory as a way of understanding the ways in which these lived experiences lead to service user and caregiver being identified as particular kinds of participant in these processes.
- (ii) Our contributions to theories of service user work and capacity that have included Normalisation Process Theory,^{45,46} and Burden of Treatment Theory¹ and participation in the development of the Cumulative Complexity Model¹⁵. These have specified particular configurations of normative expectations of service users and caregivers (negotiated and assumed obligations), patterns of workload and capacity and their consequences.
- (iii) Structural theories of social inequalities,⁴⁷ networks,⁴⁸ and social capital.⁴⁹ These provide a set of fundamental conceptual building blocks for modelling important the social context of burden of treatment for service users and caregivers.

We will draw together key concepts from these theoretical frameworks in a taxonomy that will form the basis of a coding manual that will define what we are looking for in the manual analysis of papers included in the review. They will also form the core of a lexicon that can be used to define semantic and relational searches in text mining.

6.2 Systematic literature searches. The protocol for the review will be made publicly available on PROSPERO. Literature searching will be contracted out to York Health

workshops impossible. With the agreement of NIHR HSDR colleagues these workshops were dropped from the study design and replaced with a series of informal discussions with individual colleagues across the health and social care field as the project develops.

Economics Consortium (YHEC), who have an internationally acknowledged team of information specialists. In collaboration with them, we will develop a search strategy for systematic searches of the following databases: Social Care Online, Science, Social Science and Arts and Humanities Citation Indices (Web of Science); CINAHL (EBSCO Host); EMBASE (Ovid); MEDLINE (Ovid); PsycINFO (Ovid); Scopus; PubMed. Search development is likely to be an iterative process given the complexity of the topic. The search will be peer reviewed by a second information specialist and the performance of the strategy in finding known relevant studies will be tested.

YHEC will run searches and conduct de-duplication of citations, providing Endnote database files of citations for screening. The search strategy is likely to use a multi-stranded approach using several different conceptual combinations, reflecting the fact that relevant records may not be consistently described. The social care literature may use different terms to describe phenomena of interest to the health care field. An example of the multi-stranded approach employed in this work might be: (1) (Index conditions OR generic terms for long term conditions) AND experience terms; (2) (Index conditions OR generic long term conditions terms) AND concept of service users AND qualitative research terms; (3) 1 OR 2; limit 3 to English language; (4) limit 4 to records including abstracts; (5) limit 5 to records published in the year 2010 onwards; (6) NOT (editorials OR comments etc.). A complementary search strategy will be developed to locate studies that pertain to informal carers. Further strands (i.e. conceptual combinations) are likely to be identified during strategy development.

6.3 Inclusion criteria. We will *include* reports that meet all of the following general criteria, but we will tailor or stratify the inclusion criteria to meet specific features of index conditions.

6.3.1 Participants. people aged >18 years; diagnosed with Parkinson's disease, ulcerative colitis, brain cancer, rapidly progressing dementia, bipolar disorder, schizophrenia, and their caregivers.

6.3.2 Reports. results of qualitative studies of service users' or carers' accounts of the lived experiences of eligible conditions; their interactions with health professionals, healthcare provider organisations, treatment settings, technologies and regimens of care and self-care; and the social and economic contexts in which experiences of illness and care are set.

6.3.3 Study designs. primary qualitative studies using semi-structured and unstructured interviews; primary qualitative studies using participant or non-participant observation studies; systematic reviews of qualitative studies, qualitative meta-syntheses and meta-ethnographies.

6.3.4 Settings. Studies of illness experiences within self-care programmes, healthcare systems, and social care systems.

6.3.5 Date of publication. Because there have been important changes in the organisation of care (and especially self-care) in recent years, we will restrict eligible studies to those published between 1 January 2010 and 31 December 2019.

6.3.6: Language. English.

6.4: Exclusion criteria. We will *exclude* reports which do not report the results of qualitative research with service users or carers; reports of treatment effectiveness, for example, RCTs, where the focus is on the treatment effect rather than the service user's or carer's experience; reports of healthcare organisation or delivery which are not focused on service user's or carer's experience; and editorials, notes, letters and case reports.

6.5 Screening. Searches are likely to generate a very large number of records and so first stage screening will eliminate those that are obviously irrelevant, such as notes, comments, editorials, non-systematic reviews, RCTs and studies in diseases that are not eligible. Second stage screening will start with an assessment of relevance of citations and abstracts by two reviewers independently. Any studies which are eligible (i.e. they meet the criteria set out above) or which may be eligible (i.e. where the content is unclear, or reviewers disagree) will be obtained in full text. If agreement about inclusion cannot be reached, we will call on an independent assessor to act as final arbiter. Full text papers will be screened by two reviewers independently. Papers selected for inclusion will be stored as .pdf files in secure Endnote Libraries with automatic back up. Records excluded based on assessment of full text will be listed in an excluded studies table with a brief reason for exclusion.

6.6 Quality assessment of eligible articles. There are many proposed sets or reporting criteria for qualitative studies. We will use the Consolidated Criteria for reporting Qualitative Research (COREQ)⁵⁰ checklist to assess the quality of qualitative research proposals and papers. It provides clear criteria for identifying high-quality reports. However, since there is no universally accepted reporting standard for qualitative studies, COREQ can only guide decision-making on eligibility for inclusion. This is especially important because we will be drawing on bodies of literature (e.g. social work and social care) that may have different disciplinary criteria for reporting. Reports that provide insufficient information about sample, question, method and setting will be excluded from the review.

6.7 Data extraction. We will use two approaches to data extraction:

6.7.1: Manual Data Extraction. We will undertake open and theory-informed coding. In theory-informed coding we will use the taxonomy of theoretical constructs described in Section 5.1.1 above. We will design a data extraction instrument, develop a coding strategy, and write a coding framework and manual.⁴² As in our earlier reviews, we will test and refine this in a preliminary analysis of a sample of papers. This coding framework will be integrated into NVivo 12® Software. Researchers will then independently read and code papers, recording the results of this work in NVIVO files. Where disagreements about coding occur, they will be arbitrated by a third member of the team.

6.7.2: Text mining. Leximancer® is a text mining application that is used in studies of consumer behaviour and marketing and in systematic reviewing in computer science and related fields.⁵¹ It has been used very little used in health services research. We will integrate our coding framework or lexicon in Leximancer's preprocessing module. We will then run this across the whole data set of included papers.⁵² Leximancer uses semantic and relational algorithms to search for frequencies of groups of terms and for associations between them,⁵¹ and it produces maps and models of the relationships between them.⁵³ It thus identifies empirical regularities in natural language data, and suggests ways in which they are connected.⁵⁴ We will investigate these: searches will be informed by terms from our own coding framework as well as its own open coding of a qualitative data set, we expect that it will suggest new concept labels, and new patterns of lexical association between them.⁵⁵

6.8 Data analysis. Qualitative data analysis will follow the three-stage process that we have previously used to develop robust conceptual model. Our approach to data analysis is abductive⁵⁶ in perspective, informed by attribution theory.⁵⁷ We are searching for different kinds of empirical regularities in natural language data, and for the ways that these include characterisation and explanation of relevant phenomena, rather than for the de novo emergent themes that would be discovered in an inductive phenomenological or grounded theory study.

6.8.1 Taxonomy-building. The first phase of analysis is descriptive. Using both manual and text mining applications we will produce taxonomies of

- (i) the work of service users and caregivers in participating and engaging with the expectations of self-care, health and social care providers;
- (ii) the work of negotiating and interacting with health and social care providers and professionals;
- (iii) the shaping effects of social inequalities and structural disadvantages.

The content of these taxonomies will vary between index conditions and disease trajectories, but they will produce large numbers of potential taxa. To prepare for modelling, we will identify and eliminate redundancies and duplicates amongst these.

6.8.2 Characterisation and modelling. We will characterise and compare patterns of taxa and constructs for six index conditions; three disease trajectories; three service contexts, and one universal set of constructs which appear in all index conditions and all disease trajectories. In the early phases of modelling, we will therefore assess the relative significance and the degree of universality of any particular construct. We will also assess the position and role of constructs in relation to each other. For example, these constructs may characterise preconditions, resources, relationships, or endpoints. We will then sift and sort constructs, writing them as context-independent propositions that are linked to four general categories of work revealed in the literature over five decades: *being* (negotiating experience and identity); *changing* (managing status passage and biographical disruption); *relating* (participating in interactions within healthcare provision and informal social networks); and *doing* (performing health behaviours and enacting healthcare technologies and self-care practices). We will continue this until we reach the most parsimonious possible model of interactions between constructs.

6.8.3 Construct validation. The final stage of analysis is to link context dependent propositions together in a summary statement that characterises and explains the operation of the model and its implications in ways that can be easily understood. The completed analysis and summary statement will be presented to a PPI reference groups in Wessex ARC and to service user and caregiver advocacy groups, and to a reference group that includes social and health care practitioners and researchers. At this stage, we will also explore scenarios around workload reduction strategies and person-centered service design for people with complex health problems. This is to ensure that the model's constructs and propositions have (i) face validity for people who experience health and social care provision, and for other practitioners and researchers in the field, and (ii) that they can inform supportive interventions that are practically workable across a range of treatment modalities, service organisation and delivery.

7. Dissemination, Outputs and anticipated Impact

The review is configured as part of the research programme of the innovation and implementation research theme in NIHR North Thames ARC (May) and the work programme of NIHR ARC Wessex (Richardson, ARC Director), and we link to professional education and development through Chew-Graham's networks developed as curriculum advisor to the RCPsych and as RCGP mental health champion. We will ensure wide dissemination of this systematic review and qualitative synthesis, and we will tailor outputs to ensure that they promote key messages for different groups of stakeholders.

7.1 RESEARCH OUTPUTS

7.1.1 Research outputs A high quality comparative qualitative evidence synthesis, conforming to PRISMA⁵⁸ reporting standards, leading to multiple peer-reviewed publications; conference and seminar presentations.

7.1.2 Digital materials Digital materials and animations tailored to meet the expectations of service users, caregivers and their advocates.

7.1.3 Policy outputs. Briefing materials and face to face presentations tailored for senior practitioners and policy makers.

7.2 ENGAGING SERVICE USERS, PROFESSIONALS AND POLICY MAKERS.

7.2.1 Practitioners, NHS and Social Care Managers. In the construct validation and translation phase of the study we will undertake dissemination and engagement activities with stakeholders to explore ways in which the constructs developed within the review might inform supportive interventions that are practically workable in reconfigurations of treatment modalities, service organisation and delivery. We will use ARC communications services to disseminate these results to NHS Integrated Care Systems, Sustainability and Transformation Partnerships, Clinical Commissioning Groups, Local Authorities, private and third Sector Service Providers in health and social care.

7.2.2 Service users, caregivers and their advocates. We will engage with service users and caregivers, and their advocates, we will work with stakeholders to co-create digital materials and animations which we will publish on the web using Instagram and Youtube, and also through our interactions with www.serviceuserrevolution.org.

7.2.3 Policy-makers. We will take full advantage of opportunities for face-to-face interaction, analog and digital media to promote the results of this review. In collaboration with our ARC communications colleagues and PublicPolicy@Southampton we will develop and implement a robust and ambitious strategy to communicate results of this work. To engage policy-makers at a national level we will work with PublicPolicy@Southampton to identify and engage with key national policy-makers and influencers, provide them with key policy briefings (in both web and hard copy form).

7.2.4 Research community. Dissemination to the research community will be through open access journal articles, conference presentations, and seminars. In addition to the final report to be published as a peer-review journal monograph in *Health Services and Delivery Research*, we will publish our protocol; report on our theoretical framework; methodological aspects of text mining in qualitative systematic reviews; and comparative models of service user and caregiver workload and capacity in physical and mental health problems. We will also propose a workload reduction model for service user and caregiver burden in complex disease trajectories.

8. PROJECT / RESEARCH TIMETABLE

See Appendix 1: Combined PRISMA Flowchart and workflow model

See Appendix 2: GANTT Chart

9. PROJECT MANAGEMENT

9.1 Chief Investigator. The project will be directed by Professor Carl May. He will have direct line management responsibility for the research fellow and responsibility for the administrative assistant. He will be responsible for the proper conduct of the study, any spending associated with it, and for timely reporting to NIHR and any other relevant agencies.

9.2 Investigator meetings. Prof May, the Research fellow, and the Co-investigators will meet virtually once a month (using Zoom videoconferencing software). Four in-person Investigator meetings will take place during the study. Investigator meetings will monitor progress against the protocol, interpret data, and contribute to the content of dissemination events, reports and publications. Project progress will be reported to the HSDR Board in the form and at the times specified in the contract, and to the Oversight Committee in the usual way.

9.3 Oversight committee. The principal and co-investigators, research fellow, and collaborators, will be guided in their work by an Oversight Committee. This will be chaired by Professor Fiona Poland (University of East Anglia & NIHR ARC East of England). Members of the oversight committee are

Professor Richard Byng (University of Plymouth, NIHR ARC South West)

Mr Andrew Chuter (PPI Representative)

Emerita Professor Linda Gask (University of Manchester)

Professor Claire Goodman (University of Hertfordshire, NIHR ARC East of England)

Professor Jill Manthorpe (King's College, London, NIHR ARC South London)

Professor Caroline Nicholson (University of Surrey)

Professor Patricia Wilson (University of Kent, NIHR RDS South East)

This committee will meet three times during the course of the work (participants may join in person, or virtually using Zoom or Skype videoconferencing software). They will provide advice on methodological, policy and practice questions as these arise during the course of the research. The Oversight Committee will receive progress reports from the Management Committee, and members will be invited to participate in project workshops.

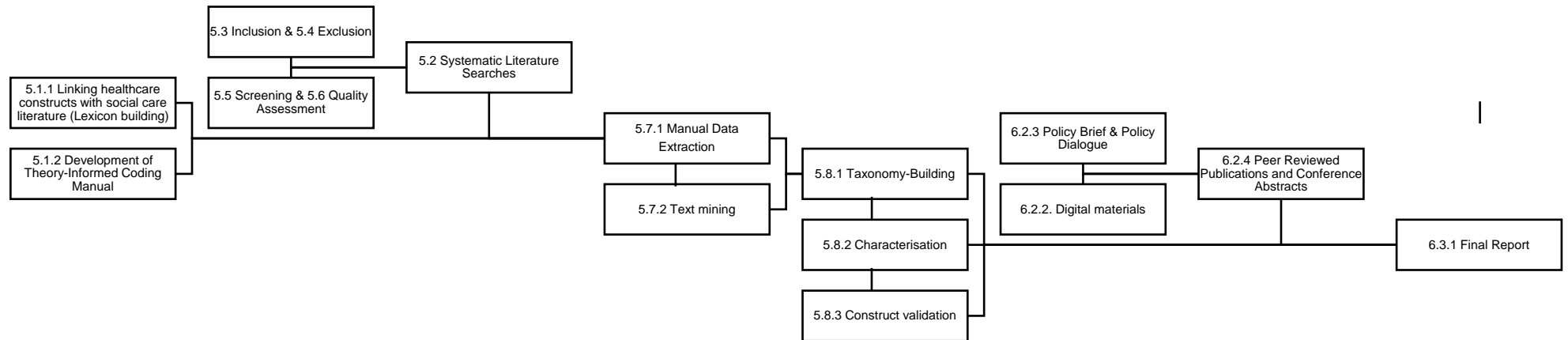
10. ETHICS / REGULATORY APPROVALS

10.1 Ethical approvals. This is a literature review and does not involve research on human subjects. Ethics Committee approval is therefore not necessary.

11. SERVICE USER AND PUBLIC INVOLVEMENT

11.1 PPI Group involvement. This proposal stems from meetings held with members of the PPI reference group of the Complexity, Service user Experience and Organisational Behaviour research theme (jointly led by May and Richardson) of NIHR CLAHRC Wessex between 2015 and 2019. Members of that group consistently pointed to the complex, time-consuming, and sometimes arduous work that they needed to do to effectively engage with NHS services. They pointed to the ways in which NHS services were often fragmented and uncoordinated, that they often experienced care pathways as arbitrary sequences of interactions, and that they struggled to make sense of the processes of care within which they were involved. In this proposed study, the lessons of that earlier PPI input have been taken on board. NIHR CLAHRC Wessex has now been superseded by NIHR ARC Wessex. Going forward we will work with the PPI group in NIHR ARC Wessex.

APPENDIX 1 Combined PRISMA Flowchart and workflow model



APPENDIX 2: GANTT CHART

Month	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24
Activity																								
5.1.1 Linking Health and Social care constructs	█	█	█	█	█	█																		
5.1.2 Coding manual development	█	█	█	█	█	█																		
5.2 Literature searches	█	█	█	█	█	█																		
5.5 & 5.6 Screening and Quality Assessment			█	█	█	█	█	█																
5.7.1. Manual Data Extraction						█	█	█	█	█	█	█	█	█										
5.7.2 Text Mining						█	█	█	█	█	█	█	█	█										
5.8.1 Taxonomy Building						█	█	█	█	█	█	█	█	█	█	█								
5.8.2 Characterisation											█	█	█	█	█	█	█	█	█					
5.8.3 Construct Validation														█	█	█	█	█	█	█	█	█	█	█
6.2. User engagement activities	█	█	█	█	█	█					█	█	█	█	█	█	█	█	█	█	█	█	█	█
6.2.2 Digital materials for service users and caregivers															█	█	█	█	█	█	█	█	█	█

REFERENCES

- 1 May, C. *et al.* Rethinking the patient: using Burden of Treatment Theory to understand the changing dynamics of illness. *Bmc Health Serv Res* **14** (1), (2014).
- 2 HEE. Framework 15: Health Education England Strategic Framework 2014-2029 (Health Education England, London, 2014).
- 3 Gately, C., Rogers, A. & Sanders, C. Re-thinking the relationship between long-term condition self-management education and the utilisation of health services. *Social Science & Medicine* **65**, 934-945 (2007).
- 4 Charmaz, K. *Good Days, Bad Days: The Self in Chronic Illness*. (Rutgers University Press, 2006).
- 5 Glaser, B. G. & Strauss, A. *Status passage*. (Routledge & Kegan Paul Ltd, 1971).
- 6 Bury, M. Chronic illness as biographical disruption. *Sociology of Health and Illness* **4**, 167-182 (1982).
- 7 Vassilev, I., Rogers, A., Kennedy, A. & Koetsenruijter, J. The influence of social networks on self-management support: a metasynthesis. *Bmc Public Health* **14**, 719 (2014).
- 8 Strauss, A., Fagerhaugh, S., Suczek, B. & Wiener, C. *The Social Organisation of Medical Work*. 2nd (1997) edn, (Transaction, 1985).
- 9 Mair, F. S. & May, C. R. *Thinking about the burden of treatment*. Vol. 349 (2014).
- 10 Gallacher, K., May, C. R., Montori, V. M. & Mair, F. S. Understanding patients' experiences of treatment burden in chronic heart failure using normalization process theory. *The Annals of Family Medicine* **9**, 235-243 (2011).
- 11 Hunt, K. J. & May, C. R. Managing expectations: cognitive authority and experienced control in complex healthcare processes. *Bmc Health Serv Res* **17**, 459, doi:10.1186/s12913-017-2366-1 (2017).
- 12 Campbell, C. Social capital, social movements and global public health: Fighting for health-enabling contexts in marginalised settings. *Soc Sci Med*, doi:10.1016/j.socscimed.2019.02.004 (2019).
- 13 McCubbin, M. Pathways to health, illness and well-being: From the perspective of power and control. *J Community Appl Soc* **11**, 75-81, doi:DOI 10.1002/casp.622 (2001).
- 14 Vassilev, I., Band, R., Kennedy, A., James, E. & Rogers, A. The role of collective efficacy in long-term condition management: A metasynthesis. *Health Soc Care Community*, doi:10.1111/hsc.12779 (2019).
- 15 Shippee, N. D., Shah, N. D., May, C. R., Mair, F. S. & Montori, V. M. Cumulative complexity: a functional, patient-centered model of patient complexity can improve research and practice. *J Clin Epidemiol* **65**, 1041-1051, doi:DOI 10.1016/j.jc1inepi.2012.05.005 (2012).
- 16 Weiss, D. *et al.* Innovative technologies and social inequalities in health: A scoping review of the literature. *Plos One* **13**, e0195447, doi:10.1371/journal.pone.0195447 (2018).
- 17 Corbin, J. & Strauss, A. Managing chronic illness at home: Three lines of work. *Qual Sociol* **8**, 224-247 (1985).
- 18 Boehmer, K. R., Abu Dabrh, A., Gionfriddo, M. R., Erwin, P. & Montori, V. M. Does the chronic care model meet the emerging needs of people living with multimorbidity? A systematic review and thematic synthesis. *Plos One* **13**, doi:ARTN e0190852 10.1371/journal.pone.0190852 (2018).
- 19 Barnett, K. *et al.* Epidemiology of multimorbidity and implications for health care, research, and medical education: a cross-sectional study. *Lancet* **380**, 37-43, doi:10.1016/S0140-6736(12)60240-2 (2012).
- 20 May, C. Chronic illness and intractability: professional-patient interactions in primary care. *Chronic Illness* **1**, 15-20 (2005).

- 21 Cornwell, J. *Hard-Earned Lives: Accounts of Health and Illness from East London*. (Routledge, 1984).
- 22 Blaxter, M. *Health and lifestyles*. (Routledge, Chapman & Hall, Inc., 1990).
- 23 Keaver, L., Perez-Ferrer, C., Jaccard, A. & Webber, L. Future trends in social inequalities in obesity in England, Wales and Scotland. *J Public Health (Oxf)*, doi:10.1093/pubmed/fdz022 (2019).
- 24 Lago, S. *et al.* Socioeconomic status, health inequalities and non-communicable diseases: a systematic review. *Z Gesundh Wiss* **26**, 1-14, doi:10.1007/s10389-017-0850-z (2018).
- 25 Boger, E. *et al.* Self-management and self-management support outcomes: A systematic review and mixed research synthesis of stakeholder views. *Plos One* **10**, e0130990 (2015).
- 26 May, C., Finch, T., Mair, F. & Mort, M. Towards a wireless patient: chronic illness, scarce care and technological innovation in the United Kingdom. **61**, 1485-1494 (2005).
- 27 Newbould, J., Taylor, D. & Bury, M. Lay-led self-management in chronic illness: a review of the evidence. *Chronic Illn* **2**, 249-261, doi:10.1179/174592006X157508 (2006).
- 28 Demain, S. *et al.* Living With, Managing and Minimising Treatment Burden in Long Term Conditions: A Systematic Review of Qualitative Research. *Plos One* **10**, e0125457 (2015).
- 29 Sav, A. *et al.* Consumer health organisations for chronic conditions: why do some people access them and others don't? *Prim Health Care Res Dev* **15**, 418-429, doi:10.1017/S1463423614000036 (2014).
- 30 Sav, A. *et al.* The ideal healthcare: priorities of people with chronic conditions and their carers. *Bmc Health Serv Res* **15**, 551, doi:10.1186/s12913-015-1215-3 (2015).
- 31 Sav, A. *et al.* Burden of treatment for chronic illness: a concept analysis and review of the literature. *Health Expect* (2013).
- 32 May, C. R. *et al.* Experiences of long-term life-limiting conditions among patients and carers: what can we learn from a meta-review of systematic reviews of qualitative studies of chronic heart failure, chronic obstructive pulmonary disease and chronic kidney disease? *BMJ Open* **6**, e011694, doi:10.1136/bmjopen-2016-011694 (2016).
- 33 May, C. Rethorizing the clinical encounter: Normalization processes and the corporate ecologies of care. *Assaults on the lifeworld: new directions in the sociology of chronic and disabling conditions*. London: Routledge, 129-145 (2010).
- 34 May, C., Montori, V. M. & Mair, F. S. We need minimally disruptive medicine. *BMJ* **339**, b2803-, doi:10.1136/bmj.b2803 (2009).
- 35 Boehmer, K. R. *et al.* Patient capacity and constraints in the experience of chronic disease: a qualitative systematic review and thematic synthesis. *Bmc Fam Pract* **17**, 127, doi:10.1186/s12875-016-0525-9 (2016).
- 36 Jani, B. *et al.* Identifying treatment burden as an important concept for end of life care in those with advanced heart failure. *Current opinion in supportive and palliative care* **7**, 3-7 (2013).
- 37 Gallacher, K. *et al.* Uncovering treatment burden as a key concept for stroke care: a systematic review of qualitative research. *Plos Med* **10**, e1001473 (2013).
- 38 Gallacher, K. *et al.* Uncovering treatment burden as a key concept for stroke care: a systematic review of qualitative research. *PLoS Med* **10**, e1001473, doi:10.1371/journal.pmed.1001473 (2013).
- 39 Lippiett, K. A., Richardson, A., Myall, M., Cummings, A. & May, C. R. Patients and informal caregivers' experiences of burden of treatment in lung cancer and chronic obstructive pulmonary disease (COPD): a systematic review and synthesis of qualitative research. *BMJ Open* **9**, e020515, doi:10.1136/bmjopen-2017-020515 (2019).
- 40 May, C. R. *et al.* EXPERTS 1—experiences of long-term life-limiting conditions among patients and carers: protocol for a qualitative meta-synthesis and conceptual modelling study. *BMJ open* **5**, e007372 (2015).

- 41 May, C. R. *et al.* Experiences of long-term lifelimiting conditions among patients and carers: what can we learn from a meta-review of systematic reviews of qualitative studies of chronic heart failure, chronic obstructive pulmonary disease and chronic kidney disease? *BMJ Open* **6**, e011694., doi:doi:10.1136/bmjopen-2016-011694 (2016).
- 42 Rapley, T. *How to study conversations, discourse and documents.* (Sage, 2006).
- 43 Corbin, J. & Strauss, A. *Unending Work and Care: Managing Chronic Illness at Home.* (Jossey-Bass, 1988).
- 44 Charmaz, K. The body, identity, and self: Adapting to impairment. *Sociological Quarterly* **36**, 657-680 (1995).
- 45 May, C. *et al.* Development of a theory of implementation and integration: Normalization Process Theory. *Implement Sci* **4**, doi:doi:10.1186/1748-5908-4-29 (2009).
- 46 May, C. in *Handbook of the Sociology of Health, Illness, and Healing: A Blueprint for the 21st Century* (eds Bernice. Pescosolido, Jack A Martin, & Anne Rogers) (Springer, 2010).
- 47 Doyal, L. *The political economy of health.* (Pluto Press Ltd, 1979).
- 48 Granovetter, M. The strength of weak ties. *Am J Sociol* **78**, 1360-1380 (1973).
- 49 Lin, N. *Social capital : a theory of social structure and action.* (Cambridge University Press, 2001).
- 50 Tong, A., Sainsbury, P. & Craig, J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care* **19**, 349 - 357 (2007).
- 51 Stockwell, P., Colomb, R. M., Smith, A. E. & Wiles, J. Use of an automatic content analysis tool: A technique for seeing both local and global scope. *Int J Hum-Comput St* **67**, 424-436, doi:10.1016/j.ijhcs.2008.12.001 (2009).
- 52 Thompson, J., Davis, J. & Mazerolle, L. A systematic method for search term selection in systematic reviews. *Research Synthesis Methods* **5**, 87-97, doi:10.1002/jrsm.1096 (2014).
- 53 Watson, M., Smith, A. & Watter, S. Leximancer concept mapping of patient case studies. *Knowledge-Based Intelligent Information and Engineering Systems, Pt 3, Proceedings* **3683**, 1232-1238 (2005).
- 54 Neal, M. R. Textual Data Collection and Analysis for Human Factors. *Adv Hum Fact Erg Ser*, 595-604 (2013).
- 55 Harwood, I. A., Gapp, R. P. & Stewart, H. J. Cross-Check for Completeness: Exploring a Novel Use of Leximancer in a Grounded Theory Study. *Qualitative Report* **20**, 1029-1044 (2015).
- 56 Timmermans, S. & Tavory, I. Theory construction in qualitative research from grounded theory to abductive analysis. *Sociol Theor* **30**, 167-186 (2012).
- 57 Fosterling, F. *Attribution.* (Psychology Press, 2001).
- 58 Moher, D., Liberati, A., Tetzlaff, J., Altman, D. G. & Group, P. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLoS Med* **6**, e1000097, doi:10.1371/journal.pmed.1000097 (2009).