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# BMJ Open

## The specification, acceptability and effectiveness of respite care and short breaks for young adults with complex healthcare needs: protocol for a mixed-methods systematic review

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**TITLE PAGE**

**Title:** The specification, acceptability and effectiveness of respite care and short breaks for young adults with complex healthcare needs: protocol for a mixed-methods systematic review

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**MANUSCRIPT**

**Title:** The specification, acceptability and effectiveness of respite care and short breaks for young adults (18-40 years) with complex healthcare needs: protocol for a mixed-methods systematic review.

**Abstract**

Introduction

The number of young adults with complex healthcare needs (CHCNs) due to life-limiting conditions/complex physical disability has risen significantly as children with complex conditions survive into adulthood. Respite care and short breaks are an essential service, however needs often go unmet after the transition to adult services, leading to a significant impact on the life expectancy and quality of life for this population. We aim to identify, appraise, and synthesise relevant evidence to explore respite care and short breaks provision for this population, and to develop a conceptual framework for understanding service models.

Methods and Analysis

A mixed-methods systematic review conducted in two stages: 1) knowledge map and 2) evidence review. We will comprehensively search multiple electronic databases; utilise the CLUSTER approach, search relevant websites, and circulate a ‘call for evidence’. Using the SPICE framework, two reviewers will independently select evidence for inclusion into a knowledge map and subsequent evidence review, extract data relating to study and population characteristics, methods, and outcomes; and assess the quality of evidence. A third reviewer will arbitrate where necessary.

Evidence will be synthesised using the following approaches: quantitative (narratively/conducting meta-analyses where appropriate); qualitative (framework approach); policy and guidelines (documentary analysis informed approach). An overall, integrated synthesis will be created using a modified framework approach. We will use GRADE/GRADE-CERQual to assess the strength and confidence of the synthesised evidence. Throughout, we will develop a conceptual framework to articulate how service models work in relation to context and setting.

## Ethics and dissemination

Ethical approval is not required as this is a systematic review. We will present our work in academic journals, at appropriate conferences; we will disseminate findings across networks using a range of media. Steering and advisory groups were established to ensure findings are shared widely and in accessible formats.

**PROSPERO registration number:** CRD42018088780

## Strengths and limitations of this study

- The first systematic review of all available evidence on the nature and provision of respite and short break services for young adults with complex healthcare needs.
- Aims and review questions were developed with stakeholders from the advisory group including young adults with complex healthcare needs, parents, and professionals.
- Use of a mixed-methods approach to enable inclusion of all types of qualitative, quantitative and policy evidence.
- Development of a knowledge map to characterise current services and a conceptual framework to inform future service provision and further research.
- There may be limited quantitative and health economic data from which to draw firm conclusions.

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**INTRODUCTION**

Young adults with life-limiting conditions (LLCs) and young adults with complex physical disabilities often live with multiple co-morbidities due to their complex healthcare needs (CHCNs). Care for these young adults is an ongoing complex process, with no simple care pathway, and often multiple, unplanned episodes of illness. The number of children with CHCNs who survive to become young adults is rising annually [1,2]. In 2010 there were 55,721 young adults with complex needs living in England [3] and an estimated 100,000 disabled children with complex care needs in England in 2007 [4]. This growing population require appropriate services to meet healthcare needs as they transition from children to adult services, including respite care and short breaks which are an essential component of support for young adults with CHCNs and their families [5,6]. Table 1 details key definitions used in this systematic review protocol.

Respite care and short breaks are beneficial to the person receiving care, their carers and families; for example increasing family carer resilience [7], improving psychological well-being of parents [5,8], reducing risk of carer breakdown [7,9], and avoiding costly unplanned hospital admissions, length of stay or social care intervention [10,11]. Inadequate provision of services for young adults transitioning to adult care has a significant impact on life expectancy and quality of life, and increases the psychosocial burden on families and carers [12–15]. Seven out of ten families who care for someone with profound or multiple disabilities have reached, or come close to, ‘breaking point’ due to lack of short break services [16].

In children services short breaks provide opportunities for children to enjoy social interaction, support for family carers, and support for siblings [17]. Examples include residential schools, sitting services, day care in the home or other settings, or packages tailored to individual needs [18]. In adult services, planned respite or replacement care focuses on support for carers rather than for the person receiving care. Typically, adult services meet the needs of older people with cancer or other terminal diagnoses, and may therefore be inappropriate for young adults with fluctuating health conditions, such as those with CHCNs [5,9,13,19,20]. Limited respite care, particularly for those with very complex healthcare

needs, is available for planned short breaks or emergency family situations once young adults with CHCNs have transitioned to adult services [7,21,22].

Despite the rising number of young people with CHCNs surviving into early adulthood and the consequent increase in service demand, the current scale, cost, and types of available respite care have not been collated and systematically evaluated. The optimum service model for the provision of respite care and short breaks is currently uncertain, therefore a systematic review of the available evidence is needed to inform the development of future services and to identify research priorities.

Table 1 Key definitions

<b>CHCNs</b>	Substantial and ongoing healthcare needs, typically across multiple health concerns, requiring a co-ordinated response from more than one service.
<b>Complex physical disability</b>	Impairments and/or physical disabilities, due to congenital or acquired physical disability, or major neurological trauma, that requires a complex level of physical management and support.
<b>LLCs</b>	A life-limiting or life-threatening condition where there is no reasonable hope of cure and from which the person is expected to die.
<b>Respite care and short breaks</b>	The temporary provision of formal (paid) or informal (unpaid) physical, emotional, spiritual or social care for a dependent person., defined as follows. Formal respite care is provided by organisations or individuals who receive financial payment, including family carers paid through management of personal care budgets. Informal respite care does not involve financial payment.

## OBJECTIVES

The aims of this systematic review are to identify, appraise, and synthesise evidence on the specification, acceptability, effectiveness and facilitators or barriers to respite care and short breaks provision for young adults with CHCNs due to a LLC or complex physical disability. The specific objectives are as follows:

1. To identify and characterise the different types of formal and informal respite care and short break provision for young adults (18-40 years) with CHCNs due to a LLC or complex physical disability.
2. To determine the effectiveness and cost-effectiveness of different types of formal and informal respite care and short break provision for young adults (18-40 years) with CHCNs due to a LLC or complex physical disability.

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3. To better understand the impact, experiences and perceptions of respite care and short break provision from the perspectives of service users and providers.
4. To explore current United Kingdom (UK) policy, not-for-profit-organisation (NFPO) publications and guideline recommendations regarding respite care and short break provision for young adults (18-40 years) with CHCNs due to a LLC or complex physical disability.
5. To develop a conceptual framework that shows the programme logic and articulates the programme theories of respite care and short break models for young adults (18-40 years) with CHCNs due to a LLC or complex physical disability that will inform service planning and commissioning.
6. To make recommendations for further empirical research to inform intervention development and evaluation.

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A PPI advisory group (PAG) of young adults and parents have supported development of the systematic review protocol, including the systematic review questions and key definitions to facilitate the process being relevant, accessible, accountable and acceptable [23,24]. The group communicates through a variety of methods to fit with the needs of individuals including face-to-face meetings, email, telephone and video communication. The PAG will continue to collaborate with the review team at key points throughout the study including contextualisation of the findings and dissemination plan [25]. The steering group includes the review team, external professionals, representatives from national stakeholder organisations and two PAG representatives, including a young adult who is the co-chair.

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Design

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The overall design is a results-based, convergent synthesis, utilising a mixed-methods systematic review design: quantitative and qualitative data will be synthesised and presented separately, with a further synthesis of the two data types undertaken to create a third, integrated synthesis [26]. The

review methods are described in accordance with guidelines (PRISMA-P) for the reporting of protocols of systematic reviews [27]. We have adopted a two-stage approach for this mixed-methods systematic review to encompass the broad review questions and facilitate stakeholder involvement, based on methods in similar evidence syntheses [28,29]. The review processes are shown in Figure 1.

The following review questions focus on young adults (18-40 years) with CHCNs:

- 1) What types of respite care and short breaks are provided in the UK and similar global economies?
- 2) What is the effectiveness and cost-effectiveness of different types of formal and informal respite care and short break provision?
- 3) What is the economic impact of respite care and short breaks?
- 4) What are service users' and providers' views of current service provision and the need for new services?
- 5) What are the facilitators and barriers to providing, implementing, using and sustaining respite care and short breaks, taking into account the different perspectives of service users, family members and providers?
- 6) What are the current UK policy and guidance recommendations for the provision of respite care and short breaks?

In Stage 1 we will identify, categorise, and describe the evidence to create a knowledge map of different service typologies of respite care and short breaks for young adults with CHCNs and to identify gaps in the evidence base. The overall principle guiding development of the Stage 1 knowledge map is to be inclusive to ensure identification of all relevant evidence.

Evidence identified in Stage 1 will be considered for inclusion in Stage 2, where we will use appropriate methods to synthesise data and consider the methodological quality of the included evidence. We will construct an evidence matrix by service typology and type of evidence (effectiveness, cost-effectiveness, experience and attitudes, and policy and guidelines). We will extract key study characteristics, assess evidence quality, and narratively synthesise information

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3 using appropriate tools and techniques. We will use the knowledge map and evidence synthesis to  
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5 develop a conceptual framework of respite care provision for young adults with CHCNs.  
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8 **Eligibility criteria**  
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11 The setting, perspective, intervention/phenomenon of interest, comparison, and evaluation (SPICE)  
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13 framework [30] underpins the overall approach to searching for and selecting relevant evidence for  
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15 inclusion, detailed in Table 2. We have selected broad criteria to reflect the diversity of service  
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17 provision and will include evidence from any study design that meets the SPICE criteria. We will  
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19 identify qualitative, quantitative, and policy-related output as defined by and reported in each study,  
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21 for any follow-up duration.  
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24 Table 2 Inclusion criteria

	Inclusion	Exclusion
Setting	Services and providers of formal respite care and/or short breaks (hospices, residential care homes, adult day services, individual providers and paid carers/family carers working in home settings, informal care from unpaid family members, holiday care).	Services and providers of care other than respite care and short breaks. Services specifically commissioned for young adults with learning disability or mental health needs.
Perspective	Young adults (18–40 years) with CHCNs due to a LLC or complex physical disability receiving respite care and/or short breaks, their parents, families, carers and/or those involved in the commissioning or delivery of their care.	Young people below the age of 18 or people older than 40 years. Young adults with learning disabilities or mental health diagnoses. Young adults who do not require respite care/short breaks.
Intervention/phenomenon of interest	Formal (paid) and informal (unpaid) respite care/short breaks.	Care other than respite care and short breaks.
Comparison	Any formal or informal respite care/short break.	Care other than respite care and short breaks.
Evaluation	Evidence from 2002–current from the 35 OECD (Organisation for Economic Cooperation and Development) countries. <b>Effectiveness:</b> Service user, family, carer, and service provider reported quantitative outcomes e.g. quality of life, well-being, health impact, stress and coping, family cohesion or satisfaction with care. <b>Cost-effectiveness:</b> Information on UK costs: evaluations of the economic impact of respite care such as quality-adjusted life year (QALY), cost per admission avoided, other measures e.g. staff grade, time, equipment and transport, to estimate relevant and relative costs for each type of care provision. <b>Experience and attitudes:</b> Concepts and themes emerging from recognised methods that capture attitudes, beliefs, preferences and	Outcomes unrelated to effectiveness, experience, or economic evidence. Unconfirmed reports and anecdotal opinion e.g. newspapers, social media, online blogs. Non-UK policy or guidelines.

<p>opinions on the provision of respite care, along with all other potential outcomes.</p> <p><b>Policy and guidelines:</b> Recommendations, directives or actions and anticipated outcomes identified in UK policy statements or guidelines.</p>	
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## Information identification

### Search sources

We will search the following electronic databases from 2002-current: ASSIA (ProQuest), British Nursing Index (NICE Evidence Services, HDAS), CINAHL (EBSCO), Cochrane Central Register of Controlled Trials (Cochrane Library), Cochrane Database of Systematic Reviews (Cochrane Library), Database of Abstracts of Reviews of Effects (Cochrane Library), EMBASE (NICE Evidence Services, HDAS), Google Scholar, HMIC (NICE Evidence Services, HDAS), Joanna Briggs Institute COnNECT+, MEDLINE (OVID), NHS Economic Evaluations Database (Cochrane Library), NIHR Journals Library, PROSPERO, PsycINFO (EBSCO), Social Care Online, TRIP database, Web Of Science (Clarivate Analytics). We will search the following trials registries: International Clinical Trials Registry Platform, EU Clinical Trials Register and Clinical Trials.gov.

We will also search for grey and unpublished literature in Open Grey and Grey Literature Report, and search charity and organisational websites (e.g. Together for Short Lives, PaedPalLit and World Health Organisation). We will use the CLUSTER approach to identify additional outputs (e.g. 'sibling' papers or 'kinship' studies) from the included evidence [31]. Finally, we will circulate a 'call for evidence' via social media channels and networks/experts identified by the team, steering group and PAG.

We will limit the searches to evidence published from 1<sup>st</sup> January 2002 due to changes in patient population, service provision and policy change over the last 15 years [3]. We will include only UK-specific evidence written in English language for the policy and guideline evidence; where feasible we will include non-English evidence for other streams. All available evidence will be included in the Stage 1 knowledge map but only evidence relevant to UK service provision will be included in Stage 2.

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Search strategy

An experienced information specialist will develop tailored search strategies with the review team, steering group and PAG; a MEDLINE search strategy will be developed using key words, free-text terms, and controlled vocabulary (Appendix 1). The MEDLINE search strategy will then be translated into other databases.

Study records

Evidence selection

Search results will be de-duplicated and uploaded to Covidence, web-based systematic review management software [32]. Two reviewers will independently screen all titles and abstracts using the inclusion criteria outlined in Table 2. For the Stage 1 knowledge map we will only use the perspective and intervention components of the SPICE criteria for evidence selection, i.e. respite care or short breaks for young adults (18-40 years) with LLCs and/or complex physical disability.

We will only include evidence from mixed populations where, a) data from young adults is reported separately to those <18 or >40 years of age and b) data from those with CHCNs, LLCs or complex physical disability are reported separately to those with other conditions.

Disagreements will be resolved through discussion and consultation within the review team. We will contact study authors for further information where eligibility is unclear. We will tabulate reasons for study exclusion and bibliographic details of evidence excluded at the full-text stage will be available on request as an electronic addendum. Results of the searching, mapping, and selection processes will be reported for both stages using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines, including a flow diagram of included/excluded evidence [33].

Data extraction

Bespoke piloted data extraction forms will be used to extract information from included evidence. In Stage 1 we will extract bibliographic, population and intervention details, using the Template for Intervention Description and Replication (TIDieR) checklist as a guide [34].

In Stage 2, two reviewers will independently extract the following information for each type of evidence (effectiveness, cost-effectiveness, evidence on experience and attitudes, and policy and guidelines):

- Publication characteristics: e.g. year, dates and country of data collection, language, source of funding
- Methods: e.g. study design, duration of follow-up
- Aims, objectives, hypotheses, target audience
- Participant characteristics: e.g. type and duration of CHCNs, inclusion/exclusion criteria, age range, gender distribution, ethnicity, number in each study group, baseline characteristics, loss to follow-up
- Types of care: e.g. care provider (formal or informal), carer status (healthcare professional or not), care setting, duration of care
- Key limitations of each item of evidence
- Description of all outcomes and their reported results.

Disagreements will be resolved through consensus and arbitration through a third reviewer where required. We will contact study authors to resolve uncertainties in study reports.

### Assessment of methodological quality

Two reviewers will independently assess the quality and methodological limitations of included evidence using appropriate tools (see Table 3), including experimental, observational, qualitative, and mixed-methods study designs, and policy/guidelines evidence [35–45]. Disagreements will be arbitrated by a third reviewer until consensus is reached.

Table 3 Methodological quality assessment tools

<b>Experimental</b>	Randomised controlled trial	Cochrane Risk of Bias (RoB) tool
	Non-randomised controlled trial	Cochrane RoB tool Effective Practice and Organisation of Care (EPOC) adaptations for different study designs
	Before and after study	Cochrane RoB tool or National Institutes of Health (NIH) tool
<b>Observational</b>	Cohort	Critical Appraisal Skills Programme (CASP) for cohort studies
	Case-control	CASP for case control studies
	Cross-sectional	Center for Evidence-Based Management (CEBMa) tool
	Interrupted time-series	Cochrane RoB (EPOC adaptation)
	Case report/case series	Center for Evidence-Based Management (CEBMa) tool
	Economic evidence	British Medical Journal Checklist for authors and peer reviewers of economic submissions
<b>Qualitative</b>	Qualitative	CASP for qualitative studies
<b>Mixed-methods</b>	Mixed-methods	Mixed Methods Appraisal Tool (MMAT)

Policy	Policy/guideline document	Appraisal of Guidelines, Research and Evaluation (AGREE II)
Other	Grey literature:	Appropriate method-specific tool for the type of evidence. If a position statement – use the Authority, Accuracy, Coverage, Objectivity, Date, Significance (AACODS) tool to assess the credibility of the source.

Data synthesis methods

In Stage 1 we will classify the evidence and consider factors such as population, timing, and location to create a knowledge map of the different service typologies of respite care. Service typologies will be determined broadly by type, eligibility criteria and target population based on the data extracted to determine how they are intended to work, what they aim to achieve, what outcomes they include and for whom (programme theory) and to describe their programme logic (i.e. components and processes in place to achieve the outcomes). Through consensus, existing knowledge and scoping searches we have identified five preliminary categories of respite and short breaks: planned residential care; day care; home-based care; emergency care; holiday care (Figure 2). The classification system may be revised following completion of the knowledge map in Stage 1.

In Stage 2 we will categorise selected evidence by service typology and evidence type (see Figure 3). We anticipate an uneven distribution of the evidence and may need to implement a sampling frame to ensure that there is a representative sample of conditions. We will summarise service characteristics and the quality of the evidence for each type of service. We anticipate considerable variability within each service typology and across each evidence stream due to the nature of respite care, research methods and reporting. In the first instance we will therefore discuss the findings for each aspect of the evidence matrix and refine the planned syntheses accordingly. We will record and report deviations from this published protocol.

Evidence of effectiveness

Data from randomised, quasi-randomised controlled trials or other intervention studies (e.g. before and after studies or observational studies) will be tabulated and synthesised narratively by service type. We anticipate that meta-analyses will not be possible due the heterogeneous nature of the

evidence. However, where appropriate we will conduct meta-analyses to estimate the effects of the intervention for each outcome, in accordance with recommendations in the Cochrane Handbook for Systematic Reviews of Interventions [35]. Where data are sufficient we will conduct sensitivity analyses based on missing data and risk of bias criteria (randomisation). Analyses will be conducted using Review Manager (RevMan) Version 5 [46].

### Evidence of cost-effectiveness

We will tabulate and narratively synthesise data derived from economic evaluations (e.g. cost-utility and cost-effectiveness, reports of care costs) and other economic evidence (e.g. cost of illness or burden of disease studies) by service type. We will consider all direct and indirect costs of respite care (e.g. direct medical and non-medical costs and productivity losses) including quality adjusted life-year (QALY), admission avoided and carer burden, taking account of the following factors: population size, service type, perspective (e.g. patient/NHS and social services/societal), price year and currency, time horizon, discount rate, and type of health related quality of life (HRQoL) instrument, where applicable.

### Evidence on experience and attitudes

We will include and narratively report the results from qualitative, quantitative and mixed-methods evidence (e.g. surveys, interviews, focus groups, observational studies, case studies, process evaluations). Quantitative data will be synthesised using the same methods as for effectiveness outcomes; qualitative data will be synthesised using framework synthesis [47]. Findings from the qualitative and quantitative syntheses will be integrated using established methods for combining mixed-methods data [48].

### Evidence from policy and guidelines

The purpose of this evidence is to create framework within which we will contextualise the included evidence. We will conduct content analysis of the evidence from relevant current UK Government policy, clinical guidelines, and NFPO literature using a documentary analysis informed approach [49]

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to tabulate the evidence based on an a priori framework, following the process outlined for textual analysis [50].

Sub-group analyses

Where possible we will conduct sub-group analyses using the PROGRESS and PROGRESS-plus frameworks (place of residence, race/ethnicity, occupation, gender, religion, education, socioeconomic status, and social capital, age, disability and sexual orientation), endorsed by the Campbell and Cochrane Equity Methods Group for systematic reviews [51]. We will explore sub-groups of interest where data permits, e.g. differences in outcomes between young adults who have transitioned from paediatric to adult services, and young adults who developed CHCNs in adulthood.

Overall synthesis

We will use the Evidence for Policy and Practice Information and Co-ordinating Centre (EPPI-Centre) framework method to integrate evidence across the evidence matrix [52,53]. Using an a priori framework we will conduct within service type and evidence stream integration of qualitative and quantitative data based on the review questions [52]. Experienced team members will lead the process to ensure that there are appropriate skills to synthesise mixed-methods evidence, and we have assigned arbitrators to mediate disagreements and uncertainties. We will consider the overall impact of methodological quality on the results by removing evidence at high risk of bias and comparing the output.

Overall assessment of evidence

Two reviewers will use the Grading of Recommendations Assessment, Development and Evaluation (GRADE) or Confidence in the Evidence from Reviews of Qualitative Research (GRADE-CERQual) [54,55] systems to assess and report strength of the evidence. GRADE assesses the following domains: risk of bias, directness of the evidence, precision of effect estimates, and risks of publication bias. GRADE-CERQual assesses methodological limitations, relevance to the review question, coherence of study findings and adequacy of the data. Results will be tabulated in summary of findings tables.

## Conceptual framework

We will develop and refine a conceptual framework of respite care and short break provision for young adults with CHCNs throughout Stages 1 and 2. The programme theory (what they aim to achieve, what outcomes they include and for whom) and programme logic (components and processes in place to achieve the outcomes) for each service type developed in Stage 1, along with findings from Stage 2 will be explored through discussion within the review team, steering group, and PAG to produce a conceptual framework. We will use Cochrane guidance [57] and examples of good practice [58–60] to guide the final programme theories and logic models for the different types of respite care and produce the final conceptual framework. The framework will contain the most important components from the stage 1 and 2 findings to ensure that the concepts and relationships are clear and useful for researchers, commissioners, service providers, and policy makers to inform future research and service development.

## REVIEW REPORTING

There are no guidelines for reporting mixed-method reviews, so we will follow Cochrane guidance to select relevant reporting elements from method-specific reporting guidelines such as PRISMA for quantitative evidence and Enhancing transparency in reporting the synthesis of qualitative research (ENTREQ) for qualitative evidence [33,56].

## ETHICS AND DISSEMINATION

As this is a systematic review of published literature, ethics approval is not required. A dissemination and pathway to impact plan has been developed in conjunction with stakeholders and the PAG. Outputs including the knowledge map, results of the review, and conceptual framework will be shared with all relevant audiences through a range of networks and using a variety of media. In addition, we will present findings at key conferences and publish in peer reviewed journals.

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**CONTRIBUTORS**

All authors (GP, KK, LB, JD, BJ, MM, CM, JN, MO, BR, AT, SS) have contributed to the development of all aspects of the protocol. The review design and methodology were developed by GP, LB, JD, BJ, MM, CM, JN, MO, BR, the chief investigator SS and project manager KK. MM and GP developed the search strategies, AT contributed to the scoping of preliminary types of respite and short breaks. This manuscript was drafted by GP, with contributions from KK, LB, JD, BJ, MM, CM, JN, MO, BR, AT, SS and all authors have critically revised the manuscript and approved the final version and act as guarantors of the review.

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**DISCLAIMER**

The views and opinions expressed therein are those of the authors and do not necessarily reflect those of the HS&DR Programme, NIHR, NHS or the Department of Health.

**COMPETING INTERESTS**

None declared.

**DATA SHARING**

This is a protocol for a systematic review, we are happy to share data once the project is complete.

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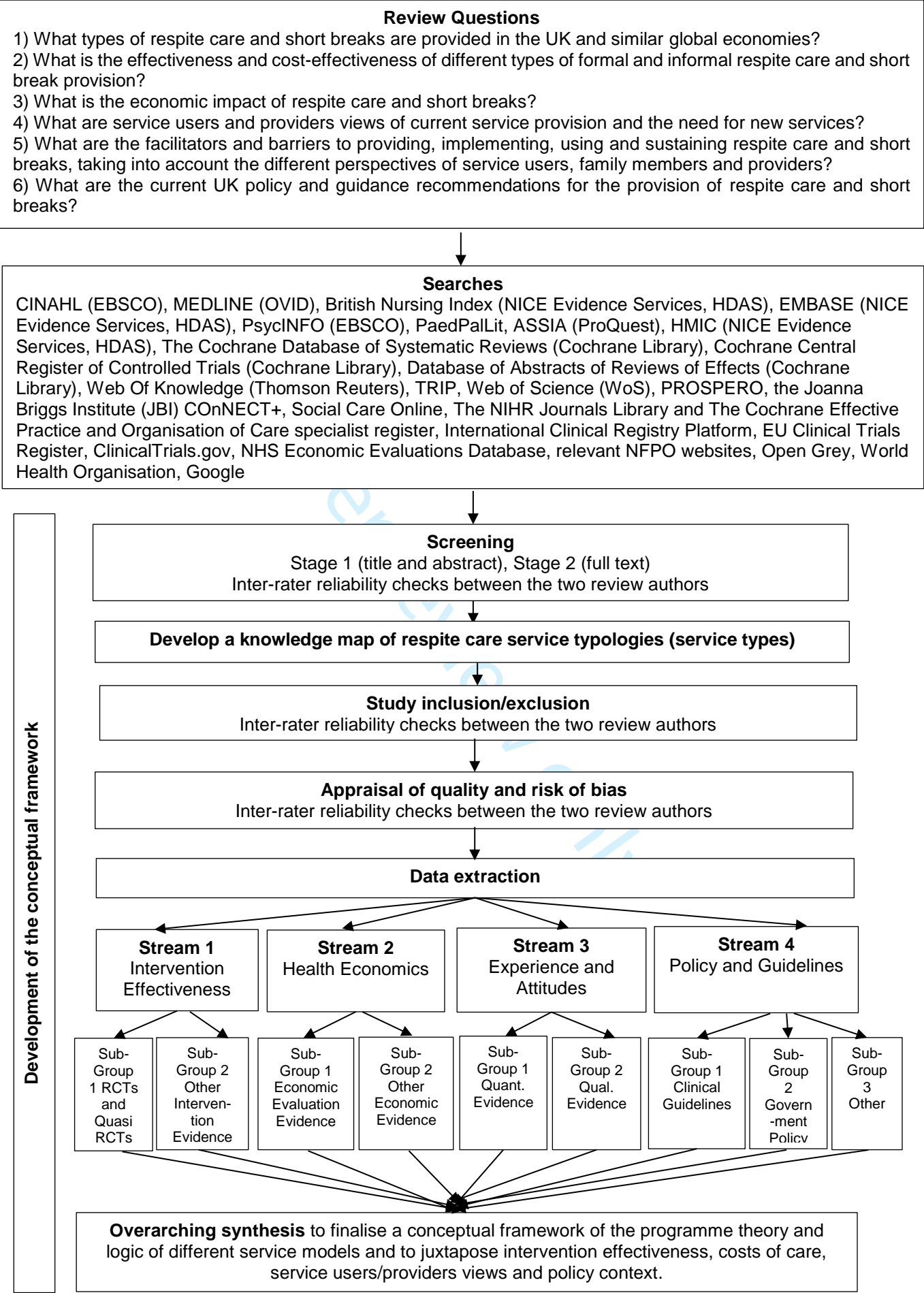
## LEGENDS

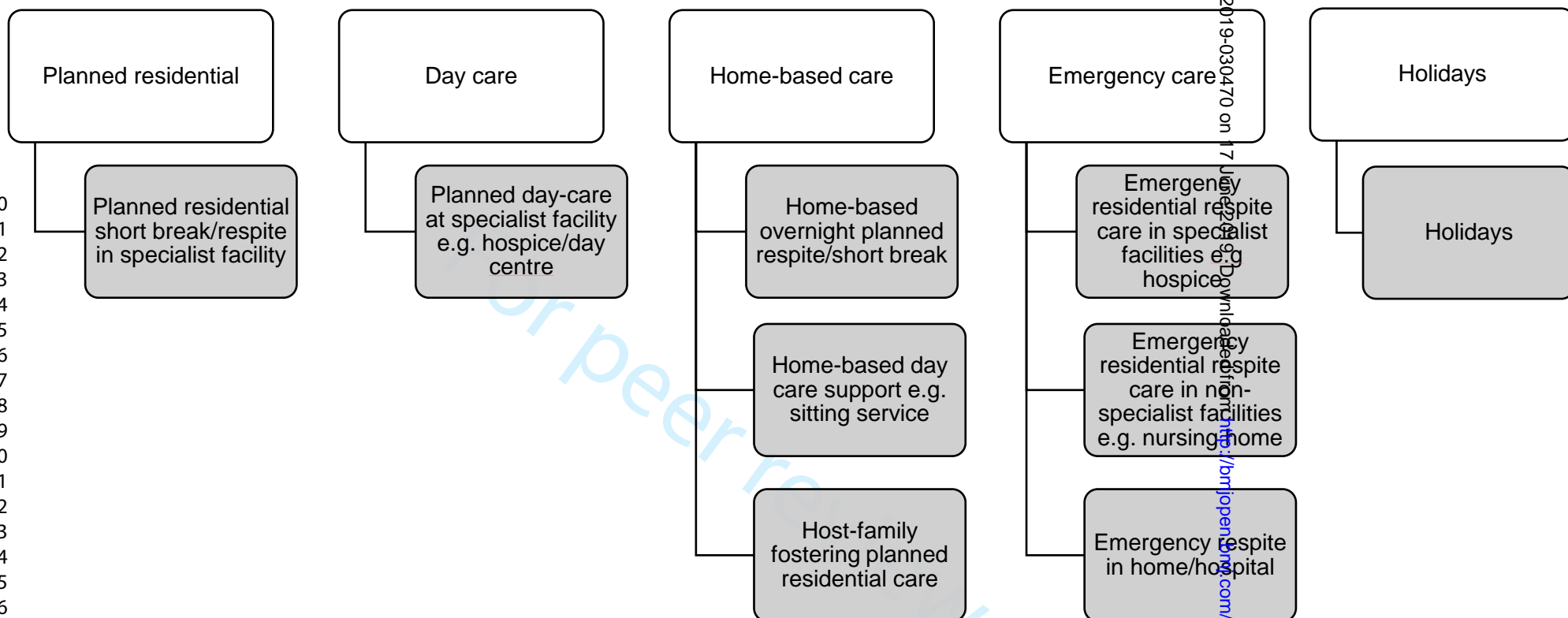
Figure 1 Review processes

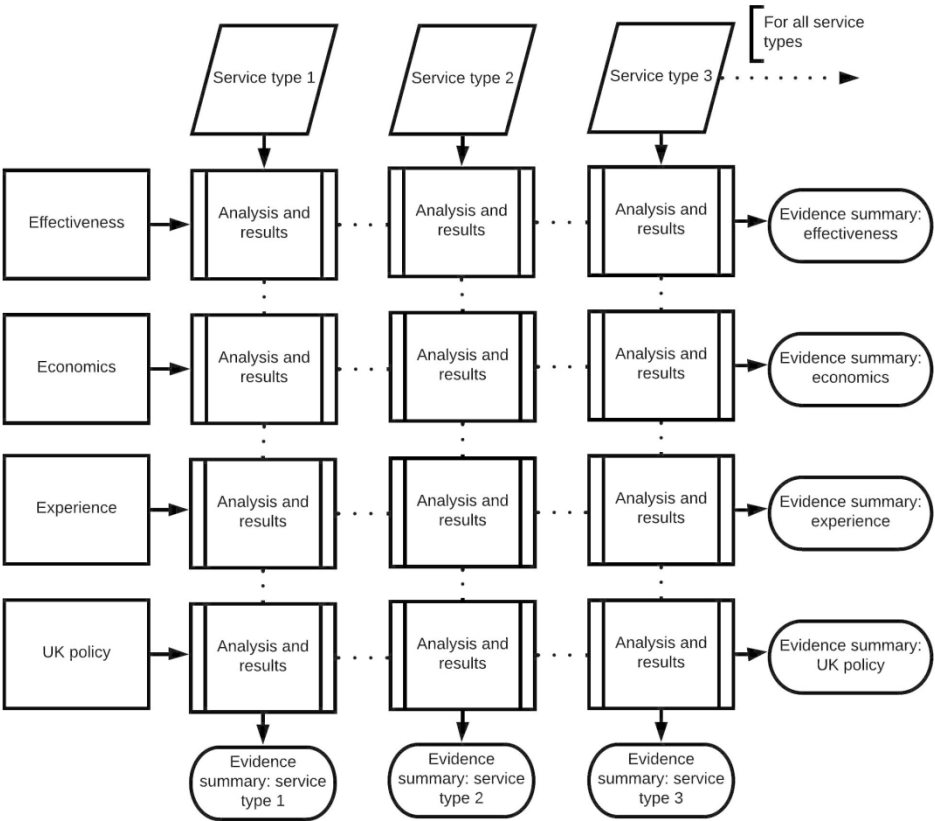
Figure 2 Initial types of respite care

Figure 3 Evidence matrix

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175x155mm (300 x 300 DPI)

**MEDLINE (Ovid)**

Access via Liverpool University

Database: Ovid MEDLINE(R) ALL &lt;1946 to September 14, 2018&gt;

Search Strategy:

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- 1 exp Respite Care/
- 2 exp Hospice Care/
- 3 exp HOSPICES/
- 4 exp "Hospice and Palliative Care Nursing"/
- 5 exp Day Care, Medical/
- 6 exp Night Care/
- 7 exp Intermediate Care Facilities/
- 8 exp Terminal Care/
- 9 exp HOLIDAYS/
- 10 "day\* away".ti,ab.
- 11 "day care\*".ti,ab.
- 12 "day centre\*".ti,ab.
- 13 "day center\*".ti,ab.
- 14 "day program\*".ti,ab.
- 15 "day service\*".ti,ab.
- 16 holiday\*.ti,ab.
- 17 "home support\*".ti,ab.
- 18 hospice\*.ti,ab.
- 19 "intermediate care".ti,ab.
- 20 "night care\*".ti,ab.
- 21 "night-time care\*".ti,ab.
- 22 "partial hospitalisation\*".ti,ab.
- 23 "partial hospitalization\*".ti,ab.
- 24 "relief care\*".ti,ab.
- 25 "relief support".ti,ab.
- 26 "residential care\*".ti,ab.
- 27 "residential home\*".ti,ab.
- 28 "residential facilit\*".ti,ab.
- 29 respite\*.ti,ab.
- 30 "short break\*".ti,ab.
- 31 "short stay\*".ti,ab.
- 32 "sitting service\*".ti,ab.
- 33 "support program\*".ti,ab.
- 34 "support scheme\*".ti,ab.
- 35 "support service\*".ti,ab.
- 36 "temporary admission\*".ti,ab.
- 37 "temporary break\*".ti,ab.
- 38 "temporary care\*".ti,ab.
- 39 "temporary relief".ti,ab.
- 40 "temporary support\*".ti,ab.
- 41 "short-term admission\*".ti,ab.
- 42 "short-term break\*".ti,ab.
- 43 "short-term care\*".ti,ab.
- 44 "short-term relief".ti,ab.
- 45 "short-term support\*".ti,ab.
- 46 "time off".ti,ab.
- 47 vacation\*.ti,ab.
- 48 "care service\*".ti,ab.
- 49 "overnight stay\*".ti,ab.
- 50 "home-based support\*".ti,ab.
- 51 "befriend\* service\*".ti,ab.
- 52 "short-break foster\*".ti,ab.
- 53 "adult placement scheme\*".ti,ab.
- 54 "shared care".ti,ab.

1 55 "replacement care".ti,ab.  
2 56 "family support".ti,ab.  
3 57 or/1-56  
4 58 exp Palliative Care/  
5 59 exp Palliative Medicine/  
6 60 exp Terminally Ill/  
7 61 exp Heart Failure/  
8 62 exp MUSCULAR DYSTROPHY, DUCHENNE/  
9 63 exp Neoplasms/  
10 64 exp Muscular Dystrophies/  
11 65 exp Cerebral Palsy/  
12 66 exp Spinal Dysraphism/  
13 67 exp Cystic Fibrosis/  
14 68 exp Disabled Persons/  
15 69 exp Disabled Children/  
16 70 exp Neurodegenerative Diseases/  
17 71 exp Multiple Trauma/  
18 72 exp Genetic Diseases, Inborn/  
19 73 exp Chromosome Disorders/  
20 74 exp "CONGENITAL, HEREDITARY, AND NEONATAL DISEASES AND  
21 ABNORMALITIES"/  
22 75 (advanc\* adj3 disease\*).ti,ab.  
23 76 (advanc\* adj3 illness\*).ti,ab.  
24 77 (advanc\* adj3 condition\*).ti,ab.  
25 78 (advanc\* adj3 disorder\*).ti,ab.  
26 79 (advanc\* adj3 abnormalit\*).ti,ab.  
27 80 (advanc\* adj3 impairment\*).ti,ab.  
28 81 (advanc\* adj3 handicap\*).ti,ab.  
29 82 (degenerative adj3 disease\*).ti,ab.  
30 83 (degenerative adj3 illness\*).ti,ab.  
31 84 (degenerative adj3 condition\*).ti,ab.  
32 85 (degenerative adj3 disorder\*).ti,ab.  
33 86 (degenerative adj3 abnormalit\*).ti,ab.  
34 87 (degenerative adj3 impairment\*).ti,ab.  
35 88 (degenerative adj3 handicap\*).ti,ab.  
36 89 (progressive adj3 disease\*).ti,ab.  
37 90 (progressive adj3 illness\*).ti,ab.  
38 91 (progressive adj3 condition\*).ti,ab.  
39 92 (progressive adj3 disorder\*).ti,ab.  
40 93 (progressive adj3 abnormalit\*).ti,ab.  
41 94 (progressive adj3 impairment\*).ti,ab.  
42 95 (progressive adj3 handicap\*).ti,ab.  
43 96 "diminished life expectancy".ti,ab.  
44 97 "limited life expectancy".ti,ab.  
45 98 cancer\*.ti,ab.  
46 99 duchenne.ti,ab.  
47 100 dying.ti,ab.  
48 101 "end of life".ti,ab.  
49 102 ("end stage renal failure" or "end stage liver failure").ti,ab.  
50 103 "heart failure".ti,ab.  
51 104 incurable.ti,ab.  
52 105 life-limit\*.ti,ab.  
53 106 "life limit\*".ti,ab.  
54 107 (life adj3 short\*).ti,ab.  
55 108 (live\* adj3 short\*).ti,ab.  
56 109 "life threaten\*".ti,ab.  
57 110 "limited life expectancy".ti,ab.  
58 111 LLC.ti,ab.  
59 112 LLI.ti,ab.

1 113 "muscular dystroph\*".ti,ab.  
2 114 neoplasm\*.ti,ab.  
3 115 "neurodegenerative condition\*".ti,ab.  
4 116 "neurodegenerative disease\*".ti,ab.  
5 117 "neurodegenerative illness\*".ti,ab.  
6 118 "neurodegenerative disorder\*".ti,ab.  
7 119 "neurodegenerative abnormalit\*".ti,ab.  
8 120 "neurodegenerative impairment\*".ti,ab.  
9 121 "neurodegenerative handicap\*".ti,ab.  
10 122 oncology.ti,ab.  
11 123 palliative.ti,ab.  
12 124 "poor prognosis".ti,ab.  
13 125 (serious\* adj3 ill\*).ti,ab.  
14 126 (terminal\* adj3 ill\*).ti,ab.  
15 127 (terminal\* adj3 care\*).ti,ab.  
16 128 (terminal\* adj3 disease\*).ti,ab.  
17 129 (terminal\* adj3 condition\*).ti,ab.  
18 130 (terminal\* adj3 disorder\*).ti,ab.  
19 131 (terminal\* adj3 abnormalit\*).ti,ab.  
20 132 (terminal\* adj3 impairment\*).ti,ab.  
21 133 (terminal\* adj3 handicap\*).ti,ab.  
22 134 (genetic adj3 disease\*).ti,ab.  
23 135 (genetic adj3 disorder\*).ti,ab.  
24 136 (genetic adj3 illness\*).ti,ab.  
25 137 (genetic adj3 condition\*).ti,ab.  
26 138 (genetic adj3 abnormalit\*).ti,ab.  
27 139 (genetic adj3 impairment\*).ti,ab.  
28 140 (genetic adj3 handicap\*).ti,ab.  
29 141 (chromosomal adj3 disease\*).ti,ab.  
30 142 (chromosomal adj3 illness\*).ti,ab.  
31 143 (chromosomal adj3 disorder\*).ti,ab.  
32 144 (chromosomal adj3 condition\*).ti,ab.  
33 145 (chromosomal adj3 abnormalit\*).ti,ab.  
34 146 (chromosomal adj3 impairment\*).ti,ab.  
35 147 (chromosomal adj3 handicap\*).ti,ab.  
36 148 (congenital adj3 disease\*).ti,ab.  
37 149 (congenital adj3 illness\*).ti,ab.  
38 150 (congenital adj3 disorder\*).ti,ab.  
39 151 (congenital adj3 condition\*).ti,ab.  
40 152 (congenital adj3 abnormalit\*).ti,ab.  
41 153 (congenital adj3 impairment\*).ti,ab.  
42 154 (congenital adj3 handicap\*).ti,ab.  
43 155 "complex health\* need\*".ti,ab.  
44 156 "early death\*".ti,ab.  
45 157 "cerebral pals\*".ti,ab.  
46 158 "spina bifida".ti,ab.  
47 159 "cystic fibrosis".ti,ab.  
48 160 encephalopath\*.ti,ab.  
49 161 disabilit\*.ti,ab.  
50 162 disabled.ti,ab.  
51 163 handicap\*.ti,ab.  
52 164 spastic\*.ti,ab.  
53 165 "impaired motor skill\*".ti,ab.  
54 166 "spinal cord condition\*".ti,ab.  
55 167 "multiple trauma".ti,ab.  
56 168 "acquired brain injur\*".ti,ab.  
57 169 "neurological condition\*".ti,ab.  
58 170 "neuromuscular condition\*".ti,ab.  
59 171 "multi-organ disease\*".ti,ab.

172 neurodisabilit\*.ti,ab.  
173 or/58-172  
174 "young adult\*".ti,ab.  
175 "young person".ti,ab.  
176 "young people".ti,ab.  
177 youth\*.ti,ab.  
178 "emerg\* adult\*".ti,ab.  
179 "early adult\*".ti,ab.  
180 (child\* adj3 transition adj3 adult\*).ti,ab.  
181 (adolescen\* adj3 transition adj3 adult\*).ti,ab.  
182 (teenage\* adj3 transition adj3 adult\*).ti,ab.  
183 (paediatric\* adj3 transition adj3 adult\*).ti,ab.  
184 (pediatric\* adj3 transition adj3 adult\*).ti,ab.  
185 "college student\*".ti,ab.  
186 "university student\*".ti,ab.  
187 "post-secondary student\*".ti,ab.  
188 undergraduate\*.ti,ab.  
189 postgraduate\*.ti,ab.  
190 exp Young Adult/  
191 exp ADOLESCENT/  
192 exp ADULT/  
193 exp FAMILY/  
194 exp CAREGIVERS/  
195 exp PARENTS/  
196 famil\*.ti,ab.  
197 carer\*.ti,ab.  
198 caregiver\*.ti,ab.  
199 parent\*.ti,ab.  
200 grandparent\*.ti,ab.  
201 relative\*.ti,ab.  
202 relation\*.ti,ab.  
203 sibling\*.ti,ab.  
204 or/174-203  
205 57 and 173 and 204  
206 limit 205 to yr="2002 -Current"

# 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	(Page No.)
ADMINISTRATIVE INFORMATION					
Title:					
Identification	1a	Identify the report as a protocol of a systematic review			1
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			4
Authors:					
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author			1
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review			17
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			17
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			5
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)			6
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			9
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			10
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			11 and Appendix 1

Study records:			
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	11-12
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)	11-12
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	13-14
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	12
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised	13-14
	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 <sup>2</sup> , Kendall's $\tau$ )	13-14
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)	15
	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)	15
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE)	15

**\* 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

## The specification, acceptability and effectiveness of respite care and short breaks for young adults with complex healthcare needs: protocol for a mixed-methods systematic review

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Secondary Subject Heading:	Research methods
Keywords:	young adult, short breaks, respite care, life-limiting condition, physical disability, systematic review

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## TITLE PAGE

**Title:** The specification, acceptability and effectiveness of respite care and short breaks for young adults with complex healthcare needs: protocol for a mixed-methods systematic review

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# MANUSCRIPT

**Title:** The specification, acceptability and effectiveness of respite care and short breaks for young adults with complex healthcare needs: protocol for a mixed-methods systematic review

## Abstract

## Introduction

The number of young adults with complex healthcare needs (CHCNs) due to life-limiting conditions/complex physical disability has risen significantly as children with complex conditions survive into adulthood. Respite care and short breaks are an essential service, however needs often go unmet after the transition to adult services, leading to a significant impact on the life expectancy and quality of life for this population. We aim to identify, appraise, and synthesise relevant evidence to explore respite care and short breaks provision for this population, and to develop a conceptual framework for understanding service models.

## Methods and Analysis

A mixed-methods systematic review conducted in two stages: 1) knowledge map and 2) evidence review. We will comprehensively search multiple electronic databases; utilise the CLUSTER approach, search relevant websites, and circulate a 'call for evidence'. Using the SPICE framework, two reviewers will independently select evidence for inclusion into a knowledge map and subsequent evidence review, extract data relating to study and population characteristics, methods, and outcomes; and assess the quality of evidence. A third reviewer will arbitrate where necessary.

Evidence will be synthesised using the following approaches: quantitative (narratively/conducting meta-analyses where appropriate); qualitative (framework approach); policy and guidelines (documentary analysis informed approach). An overall, integrated synthesis will be created using a modified framework approach. We will use GRADE/GRADE-CERQual to assess the strength and confidence of the synthesised evidence. Throughout, we will develop a conceptual framework to articulate how service models work in relation to context and setting.

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Ethics and dissemination

Ethical approval is not required as this is a systematic review. We will present our work in academic journals, at appropriate conferences; we will disseminate findings across networks using a range of media. Steering and advisory groups were established to ensure findings are shared widely and in accessible formats.

**PROSPERO registration number:** CRD42018088780

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Strengths and limitations of this study

- The first systematic review of all available evidence on the nature and provision of respite and short break services for young adults with complex healthcare needs.
- Aims and review questions were developed with stakeholders from the advisory group including young adults with complex healthcare needs, parents, and professionals.
- Use of a mixed-methods approach to enable inclusion of all types of qualitative, quantitative and policy evidence.
- Development of a knowledge map to characterise current services and a conceptual framework to inform future service provision and further research.
- There may be limited quantitative and health economic data from which to draw firm conclusions.

## INTRODUCTION

Young adults with life-limiting conditions (LLCs) and young adults with complex physical disabilities often live with multiple co-morbidities due to their complex healthcare needs (CHCNs). Care for these young adults is an ongoing complex process, with no simple care pathway, and often multiple, unplanned episodes of illness. The number of children with CHCNs who survive to become young adults is rising annually [1,2]. In 2010 there were 55,721 young adults with complex needs living in England [3] and an estimated 100,000 disabled children with complex care needs in England in 2007 [4]. This growing population require appropriate services to meet healthcare needs as they transition from children to adult services, including respite care and short breaks which are an essential component of support for young adults with CHCNs and their families [5,6]. Table 1 details key definitions used in this systematic review protocol.

Respite care and short breaks are beneficial to the person receiving care, their carers and families; for example increasing family carer resilience [7], improving psychological well-being of parents [5,8], reducing risk of carer breakdown [7,9], and avoiding costly unplanned hospital admissions, length of stay or social care intervention [10,11]. Inadequate provision of services for young adults transitioning to adult care has a significant impact on life expectancy and quality of life, and increases the psychosocial burden on families and carers [12–15]. Seven out of ten families who care for someone with profound or multiple disabilities have reached, or come close to, 'breaking point' due to lack of short break services [16].

In children services short breaks provide opportunities for children to enjoy social interaction, support for family carers, and support for siblings [17]. Examples include residential schools, sitting services, day care in the home or other settings, or packages tailored to individual needs [18]. In adult services, planned respite or replacement care focuses on support for carers rather than for the person receiving care. Typically, adult services meet the needs of older people with cancer or other terminal diagnoses, and may therefore be inappropriate for young adults with fluctuating health conditions, such as those with CHCNs [5,9,13,19,20]. Limited respite care, particularly for those with very complex healthcare

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3 needs, is available for planned short breaks or emergency family situations once young adults with  
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5 CHCNs have transitioned to adult services [7,21,22].  
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8 Despite the rising number of young people with CHCNs surviving into early adulthood and the  
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10 consequent increase in service demand, the current scale, cost, and types of available respite care  
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12 have not been collated and systematically evaluated. The optimum service model for the provision of  
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14 respite care and short breaks is currently uncertain, therefore a systematic review of the available  
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16 evidence is needed to inform the development of future services and to identify research priorities.  
17

18  
19 Table 1 Key definitions

21 22 23 <b>CHCNs</b>	Substantial and ongoing healthcare needs, typically across multiple health concerns, requiring a co-ordinated response from more than one service.
24 25 26 <b>Complex physical disability</b>	Impairments and/or physical disabilities, due to congenital or acquired physical disability, or major neurological trauma, that requires a complex level of physical management and support.
27 28 <b>LLCs</b>	A life-limiting or life-threatening condition where there is no reasonable hope of cure and from which the person is expected to die.
29 30 31 32 33 34 <b>Respite care and short breaks</b>	The temporary provision of formal (paid) or informal (unpaid) physical, emotional, spiritual or social care for a dependent person., defined as follows. Formal respite care is provided by organisations or individuals who receive financial payment, including family carers paid through management of personal care budgets. Informal respite care does not involve financial payment.

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38 **OBJECTIVES**

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41 The aims of this systematic review are to identify, appraise, and synthesise evidence on the  
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43 specification, acceptability, effectiveness and facilitators or barriers to respite care and short breaks  
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45 provision for young adults with CHCNs due to a LLC or complex physical disability. The specific  
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47 objectives are as follows:  
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- 49  
50 1. To identify and characterise the different types of formal and informal respite care and short break  
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52 provision for young adults (18-40 years) with CHCNs due to a LLC or complex physical disability.  
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55 2. To determine the effectiveness and cost-effectiveness of different types of formal and informal  
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57 respite care and short break provision for young adults (18-40 years) with CHCNs due to a LLC or  
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59 complex physical disability.  
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3. To better understand the impact, experiences and perceptions of respite care and short break provision from the perspectives of service users and providers.
4. To explore current United Kingdom (UK) policy, not-for-profit-organisation (NFPO) publications and guideline recommendations regarding respite care and short break provision for young adults (18-40 years) with CHCNs due to a LLC or complex physical disability.
5. To develop a conceptual framework that shows the programme logic and articulates the programme theories of respite care and short break models for young adults (18-40 years) with CHCNs due to a LLC or complex physical disability that will inform service planning and commissioning.
6. To make recommendations for further empirical research to inform intervention development and evaluation.

## METHODS

### Patient and public involvement (PPI)

A PPI advisory group (PAG) of young adults and parents have supported development of the systematic review protocol, including the systematic review questions and key definitions to facilitate the process being relevant, accessible, accountable and acceptable [23,24]. The group communicates through a variety of methods to fit with the needs of individuals including face-to-face meetings, email, telephone and video communication. The PAG will continue to collaborate with the review team at key points throughout the study including contextualisation of the findings and dissemination plan [25]. The steering group includes the review team, external professionals, representatives from national stakeholder organisations and two PAG representatives, including a young adult who is the co-chair.

### Design

The overall design is a results-based, convergent synthesis, utilising a mixed-methods systematic review design: quantitative and qualitative data will be synthesised and presented separately, with a further synthesis of the two data types undertaken to create a third, integrated synthesis [26]. The

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review methods are described in accordance with guidelines (PRISMA-P) for the reporting of protocols of systematic reviews [27]. We have adopted a two-stage approach for this mixed-methods systematic review to encompass the broad review questions and facilitate stakeholder involvement, based on methods in similar evidence syntheses [28,29]. The review processes are shown in Figure 1.

The following review questions focus on young adults (18-40 years) with CHCNs:

- 1) What types of respite care and short breaks are provided in the UK and similar global economies?
- 2) What is the effectiveness and cost-effectiveness of different types of formal and informal respite care and short break provision?
- 3) What is the economic impact of respite care and short breaks?
- 4) What are service users' and providers' views of current service provision and the need for new services?
- 5) What are the facilitators and barriers to providing, implementing, using and sustaining respite care and short breaks, taking into account the different perspectives of service users, family members and providers?
- 6) What are the current UK policy and guidance recommendations for the provision of respite care and short breaks?

In Stage 1 we will identify, categorise, and describe the evidence to create a knowledge map of different service typologies of respite care and short breaks for young adults with CHCNs and to identify gaps in the evidence base. The overall principle guiding development of the Stage 1 knowledge map is to be inclusive to ensure identification of all relevant evidence.

Evidence identified in Stage 1 will be considered for inclusion in Stage 2, where we will use appropriate methods to synthesise data and consider the methodological quality of the included evidence. We will construct an evidence matrix by service typology and type of evidence (effectiveness, cost-effectiveness, experience and attitudes, and policy and guidelines). We will extract key study characteristics, assess evidence quality, and narratively synthesise information

using appropriate tools and techniques. We will use the knowledge map and evidence synthesis to develop a conceptual framework of respite care provision for young adults with CHCNs.

## Eligibility criteria

The setting, perspective, intervention/phenomenon of interest, comparison, and evaluation (SPICE) framework [30] underpins the overall approach to searching for and selecting relevant evidence for inclusion, detailed in Table 2. We have selected broad criteria to reflect the diversity of service provision and will include evidence from any study design that meets the SPICE criteria. We will identify qualitative, quantitative, and policy-related output as defined by and reported in each study, for any follow-up duration.

Table 2 Inclusion criteria

	Inclusion	Exclusion
<b>Setting</b>	Services and providers of formal respite care and/or short breaks (hospices, residential care homes, adult day services, individual providers and paid carers/family carers working in home settings, informal care from unpaid family members, holiday care).	Services and providers of care other than respite care and short breaks. Services specifically commissioned for young adults with learning disability or mental health needs.
<b>Perspective</b>	Young adults (18–40 years) with CHCNs due to a LLC or complex physical disability receiving respite care and/or short breaks, their parents, families, carers and/or those involved in the commissioning or delivery of their care.	Young people below the age of 18 or people older than 40 years. Young adults with learning disabilities or mental health diagnoses. Young adults who do not require respite care/short breaks.
<b>Intervention/phenomenon of interest</b>	Formal (paid) and informal (unpaid) respite care/short breaks.	Care other than respite care and short breaks.
<b>Comparison</b>	Any formal or informal respite care/short break.	Care other than respite care and short breaks.
<b>Evaluation</b>	Evidence from 2002–current from the 35 OECD (Organisation for Economic Cooperation and Development) countries. <b>Effectiveness:</b> Service user, family, carer, and service provider reported quantitative outcomes e.g. quality of life, well-being, health impact, stress and coping, family cohesion or satisfaction with care. <b>Cost-effectiveness:</b> Information on UK costs: evaluations of the economic impact of respite care such as quality-adjusted life year (QALY), cost per admission avoided, other measures e.g. staff grade, time, equipment and transport, to estimate relevant and relative costs for each type of care provision. <b>Experience and attitudes:</b> Concepts and themes emerging from recognised methods that capture attitudes, beliefs, preferences and	Outcomes unrelated to effectiveness, experience, or economic evidence. Unconfirmed reports and anecdotal opinion e.g. newspapers, social media, online blogs. Non-UK policy or guidelines.

	opinions on the provision of respite care, along with all other potential outcomes. <b>Policy and guidelines:</b> Recommendations, directives or actions and anticipated outcomes identified in UK policy statements or guidelines.	
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Information identification

Search sources

We will search the following electronic databases from 2002-current: ASSIA (ProQuest), British Nursing Index (NICE Evidence Services, HDAS), CINAHL (EBSCO), Cochrane Central Register of Controlled Trials (Cochrane Library), Cochrane Database of Systematic Reviews (Cochrane Library), Database of Abstracts of Reviews of Effects (Cochrane Library), EMBASE (NICE Evidence Services, HDAS), Google Scholar, HMIC (NICE Evidence Services, HDAS), Joanna Briggs Institute COnNECT+, MEDLINE (OVID), NHS Economic Evaluations Database (Cochrane Library), NIHR Journals Library, PROSPERO, PsycINFO (EBSCO), Social Care Online, TRIP database, Web Of Science (Clarivate Analytics). We will search the following trials registries: International Clinical Trials Registry Platform, EU Clinical Trials Register and Clinical Trials.gov.

We will also search for grey and unpublished literature in Open Grey and Grey Literature Report, and search charity and organisational websites (e.g. Together for Short Lives, PaedPalLit and World Health Organisation). We will use the CLUSTER approach to identify additional outputs (e.g. 'sibling' papers or 'kinship' studies) from the included evidence [31]. Finally, we will circulate a 'call for evidence' via social media channels and networks/experts identified by the team, steering group and PAG.

We will limit the searches to evidence published from 1<sup>st</sup> January 2002 due to changes in patient population, service provision and policy change over the last 15 years [3]. We will include only UK-specific evidence written in English language for the policy and guideline evidence; where feasible we will include non-English evidence for other streams. All available evidence will be included in the Stage 1 knowledge map but only evidence relevant to UK service provision will be included in Stage 2.

## Search strategy

An experienced information specialist will develop tailored search strategies with the review team, steering group and PAG; a MEDLINE search strategy will be developed using key words, free-text terms, and controlled vocabulary (Appendix 1). The MEDLINE search strategy will then be translated into other databases.

## Study records

### Evidence selection

Search results will be de-duplicated and uploaded to Covidence, web-based systematic review management software [32]. Two reviewers will independently screen all titles and abstracts using the inclusion criteria outlined in Table 2. For the Stage 1 knowledge map we will only use the perspective and intervention components of the SPICE criteria for evidence selection, i.e. respite care or short breaks for young adults (18-40 years) with LLCs and/or complex physical disability.

We will only include evidence from mixed populations where, a) data from young adults is reported separately to those <18 or >40 years of age and b) data from those with CHCNs, LLCs or complex physical disability are reported separately to those with other conditions.

Disagreements will be resolved through discussion and consultation within the review team. We will contact study authors for further information where eligibility is unclear. We will tabulate reasons for study exclusion and bibliographic details of evidence excluded at the full-text stage will be available on request as an electronic addendum. Results of the searching, mapping, and selection processes will be reported for both stages using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines, including a flow diagram of included/excluded evidence [33].

### Data extraction

Bespoke piloted data extraction forms will be used to extract information from included evidence. In Stage 1 we will extract bibliographic, population and intervention details, using the Template for Intervention Description and Replication (TIDieR) checklist as a guide [34].

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3 In Stage 2, two reviewers will independently extract the following information for each type of evidence  
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5 (effectiveness, cost-effectiveness, evidence on experience and attitudes, and policy and guidelines):  
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  - Publication characteristics: e.g. year, dates and country of data collection, language, source of funding
  - Methods: e.g. study design, duration of follow-up
  - Aims, objectives, hypotheses, target audience
  - Participant characteristics: e.g. type and duration of CHCNs, inclusion/exclusion criteria, age range, gender distribution, ethnicity, number in each study group, baseline characteristics, loss to follow-up
  - Types of care: e.g. care provider (formal or informal), carer status (healthcare professional or not), care setting, duration of care
  - Key limitations of each item of evidence
  - Description of all outcomes and their reported results.

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26 Disagreements will be resolved through consensus and arbitration through a third reviewer where  
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28 required. We will contact study authors to resolve uncertainties in study reports.  
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31 **Assessment of methodological quality**  
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34 Two reviewers will independently assess the quality and methodological limitations of included  
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36 evidence using appropriate tools (see Table 3), including experimental, observational, qualitative, and  
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38 mixed-methods study designs, and policy/guidelines evidence [35–45]. Disagreements will be  
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40 arbitrated by a third reviewer until consensus is reached.  
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43 **Table 3 Methodological quality assessment tools**

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<b>Experimental</b>	Randomised controlled trial	Cochrane Risk of Bias (RoB) tool
	Non-randomised controlled trial	Cochrane RoB tool Effective Practice and Organisation of Care (EPOC) adaptations for different study designs
	Before and after study	Cochrane RoB tool or National Institutes of Health (NIH) tool
<b>Observational</b>	Cohort	Critical Appraisal Skills Programme (CASP) for cohort studies
	Case-control	CASP for case control studies
	Cross-sectional	Center for Evidence-Based Management (CEBMa) tool
	Interrupted time-series	Cochrane RoB (EPOC adaptation)
	Case report/case series	Center for Evidence-Based Management (CEBMa) tool
	Economic evidence	British Medical Journal Checklist for authors and peer reviewers of economic submissions
<b>Qualitative</b>	Qualitative	CASP for qualitative studies
<b>Mixed-methods</b>	Mixed-methods	Mixed Methods Appraisal Tool (MMAT)

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<b>Policy</b>	Policy/guideline document	Appraisal of Guidelines, Research and Evaluation (AGREE II)
<b>Other</b>	Grey literature:	Appropriate method-specific tool for the type of evidence. If a position statement – use the Authority, Accuracy, Coverage, Objectivity, Date, Significance (AACODS) tool to assess the credibility of the source.

## Data synthesis methods

In Stage 1 we will classify the evidence and consider factors such as population, timing, and location to create a knowledge map of the different service typologies of respite care. Service typologies will be determined broadly by type, eligibility criteria and target population based on the data extracted to determine how they are intended to work, what they aim to achieve, what outcomes they include and for whom (programme theory) and to describe their programme logic (i.e. components and processes in place to achieve the outcomes). Through consensus, existing knowledge and scoping searches we have identified five preliminary categories of respite and short breaks: planned residential care; day care; home-based care; emergency care; holiday care (Figure 2). The classification system may be revised following completion of the knowledge map in Stage 1.

In Stage 2 we will categorise selected evidence by service typology and evidence type (see Figure 3). We anticipate an uneven distribution of the evidence and may need to implement a sampling frame to ensure that there is a representative sample of conditions. We will summarise service characteristics and the quality of the evidence for each type of service. We anticipate considerable variability within each service typology and across each evidence stream due to the nature of respite care, research methods and reporting. In the first instance we will therefore discuss the findings for each aspect of the evidence matrix and refine the planned syntheses accordingly. We will record and report deviations from this published protocol.

## Evidence of effectiveness

Data from randomised, quasi-randomised controlled trials or other intervention studies (e.g. before and after studies or observational studies) will be tabulated and synthesised narratively by service type. We anticipate that meta-analyses will not be possible due the heterogeneous nature of the

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evidence. However, where appropriate we will conduct meta-analyses to estimate the effects of the intervention for each outcome, in accordance with recommendations in the Cochrane Handbook for Systematic Reviews of Interventions [35]. Where data are sufficient we will conduct sensitivity analyses based on missing data and risk of bias criteria (randomisation). Analyses will be conducted using Review Manager (RevMan) Version 5 [46].

Evidence of cost-effectiveness

We will tabulate and narratively synthesise data derived from economic evaluations (e.g. cost-utility and cost-effectiveness, reports of care costs) and other economic evidence (e.g. cost of illness or burden of disease studies) by service type. We will consider all direct and indirect costs of respite care (e.g. direct medical and non-medical costs and productivity losses) including quality adjusted life-year (QALY), admission avoided and carer burden, taking account of the following factors: population size, service type, perspective (e.g. patient/NHS and social services/societal), price year and currency, time horizon, discount rate, and type of health related quality of life (HRQoL) instrument, where applicable.

Evidence on experience and attitudes

We will include and narratively report the results from qualitative, quantitative and mixed-methods evidence (e.g. surveys, interviews, focus groups, observational studies, case studies, process evaluations). Quantitative data will be synthesised using the same methods as for effectiveness outcomes; qualitative data will be synthesised using framework synthesis [47]. Findings from the qualitative and qualitative syntheses will be integrated using established methods for combining mixed-methods data [48].

Evidence from policy and guidelines

The purpose of this evidence is to create framework within which we will contextualise the included evidence. We will conduct content analysis of the evidence from relevant current UK Government policy, clinical guidelines, and NFPO literature using a documentary analysis informed approach [49]

to tabulate the evidence based on an a priori framework, following the process outlined for textual analysis [50].

### Sub-group analyses

Where possible we will conduct sub-group analyses using the PROGRESS and PROGRESS-plus frameworks (place of residence, race/ethnicity, occupation, gender, religion, education, socioeconomic status, and social capital, age, disability and sexual orientation), endorsed by the Campbell and Cochrane Equity Methods Group for systematic reviews [51]. We will explore sub-groups of interest where data permits, e.g. differences in outcomes between young adults who have transitioned from paediatric to adult services, and young adults who developed CHCNs in adulthood.

### Overall synthesis

We will use the Evidence for Policy and Practice Information and Co-ordinating Centre (EPPI-Centre) framework method to integrate evidence across the evidence matrix [52,53]. Using an a priori framework we will conduct within service type and evidence stream integration of qualitative and quantitative data based on the review questions [52]. Experienced team members will lead the process to ensure that there are appropriate skills to synthesise mixed-methods evidence, and we have assigned arbitrators to mediate disagreements and uncertainties. We will consider the overall impact of methodological quality on the results by removing evidence at high risk of bias and comparing the output.

### Overall assessment of evidence

Two reviewers will use the Grading of Recommendations Assessment, Development and Evaluation (GRADE) or Confidence in the Evidence from Reviews of Qualitative Research (GRADE-CERQual) [54,55] systems to assess and report strength of the evidence. GRADE assesses the following domains: risk of bias, directness of the evidence, precision of effect estimates, and risks of publication bias. GRADE-CERQual assesses methodological limitations, relevance to the review question, coherence of study findings and adequacy of the data. Results will be tabulated in summary of findings tables.

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**Conceptual framework**

We will develop and refine a conceptual framework of respite care and short break provision for young adults with CHCNs throughout Stages 1 and 2. The programme theory (what they aim to achieve, what outcomes they include and for whom) and programme logic (components and processes in place to achieve the outcomes) for each service type developed in Stage 1, along with findings from Stage 2 will be explored through discussion within the review team, steering group, and PAG to produce a conceptual framework. We will use Cochrane guidance [56] and examples of good practice [57–59] to guide the final programme theories and logic models for the different types of respite care and produce the final conceptual framework. The framework will contain the most important components from the stage 1 and 2 findings to ensure that the concepts and relationships are clear and useful for researchers, commissioners, service providers, and policy makers to inform future research and service development.

**REVIEW REPORTING**

There are no guidelines for reporting mixed-method reviews, so we will follow Cochrane guidance to select relevant reporting elements from method-specific reporting guidelines such as PRISMA for quantitative evidence and Enhancing transparency in reporting the synthesis of qualitative research (ENTREQ) for qualitative evidence [33,60].

**ETHICS AND DISSEMINATION**

As this is a systematic review of published literature, ethics approval is not required. A dissemination and pathway to impact plan has been developed in conjunction with stakeholders and the PAG. Outputs including the knowledge map, results of the review, and conceptual framework will be shared with all relevant audiences through a range of networks and using a variety of media. In addition, we will present findings at key conferences and publish in peer reviewed journals.

## CONTRIBUTORS

All authors (GP, KK, LB, JD, BJ, MM, CM, JN, MO, BR, AT, SS) have contributed to the development of all aspects of the protocol. The review design and methodology were developed by GP, LB, JD, BJ, MM, CM, JN, MO, BR, the chief investigator SS and project manager KK. MM and GP developed the search strategies, AT contributed to the scoping of preliminary types of respite and short breaks. This manuscript was drafted by GP, with contributions from KK, LB, JD, BJ, MM, CM, JN, MO, BR, AT, SS and all authors have critically revised the manuscript and approved the final version and act as guarantors of the review.

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## DISCLAIMER

The views and opinions expressed therein are those of the authors and do not necessarily reflect those of the HS&DR Programme, NIHR, NHS or the Department of Health.

## COMPETING INTERESTS

None declared.

## DATA SHARING

This is a protocol for a systematic review, we are happy to share data once the project is complete.

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**LEGENDS**

Figure 1 Review processes

Figure 2 Initial types of respite care

Figure 3 Evidence matrix

### Review Questions

- 1) What types of respite care and short breaks are provided in the UK and similar global economies?
- 2) What is the effectiveness and cost-effectiveness of different types of formal and informal respite care and short break provision?
- 3) What is the economic impact of respite care and short breaks?
- 4) What are service users and providers views of current service provision and the need for new services?
- 5) What are the facilitators and barriers to providing, implementing, using and sustaining respite care and short breaks, taking into account the different perspectives of service users, family members and providers?
- 6) What are the current UK policy and guidance recommendations for the provision of respite care and short breaks?

### Searches

CINAHL (EBSCO), MEDLINE (OVID), British Nursing Index (NICE Evidence Services, HDAS), EMBASE (NICE Evidence Services, HDAS), PsycINFO (EBSCO), PaedPalLit, ASSIA (ProQuest), HMIC (NICE Evidence Services, HDAS), The Cochrane Database of Systematic Reviews (Cochrane Library), Cochrane Central Register of Controlled Trials (Cochrane Library), Database of Abstracts of Reviews of Effects (Cochrane Library), Web Of Knowledge (Thomson Reuters), TRIP, Web of Science (WoS), PROSPERO, the Joanna Briggs Institute (JBI) CoNNECT+, Social Care Online, The NIHR Journals Library and The Cochrane Effective Practice and Organisation of Care specialist register, International Clinical Registry Platform, EU Clinical Trials Register, ClinicalTrials.gov, NHS Economic Evaluations Database, relevant NFPO websites, Open Grey, World Health Organisation, Google

### Screening

Stage 1 (title and abstract), Stage 2 (full text)  
Inter-rater reliability checks between the two review authors

### Develop a knowledge map of respite care service typologies (service types)

### Study inclusion/exclusion

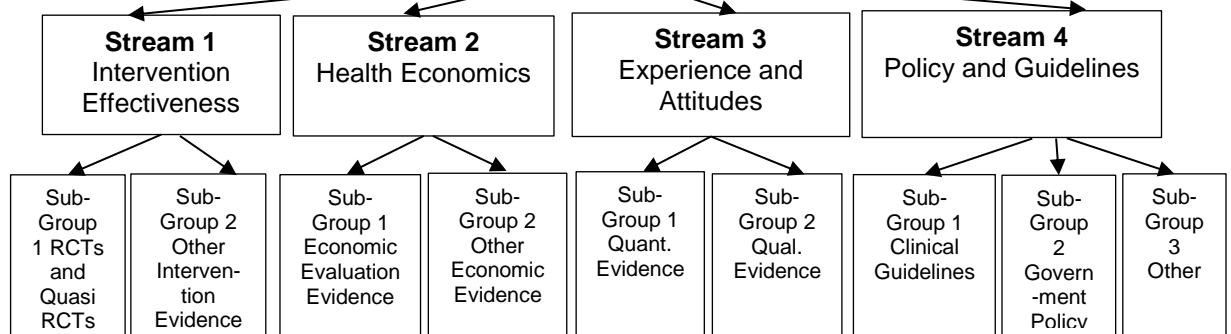
Inter-rater reliability checks between the two review authors

### Appraisal of quality and risk of bias

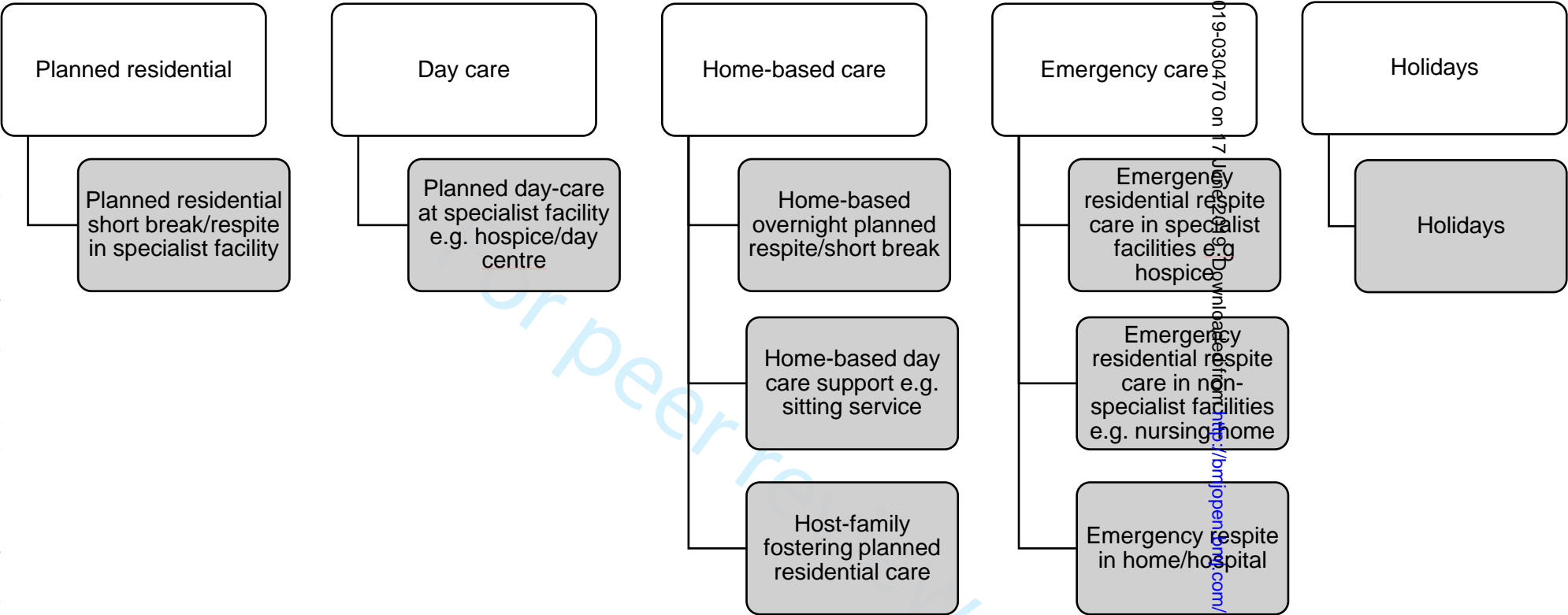
Inter-rater reliability checks between the two review authors

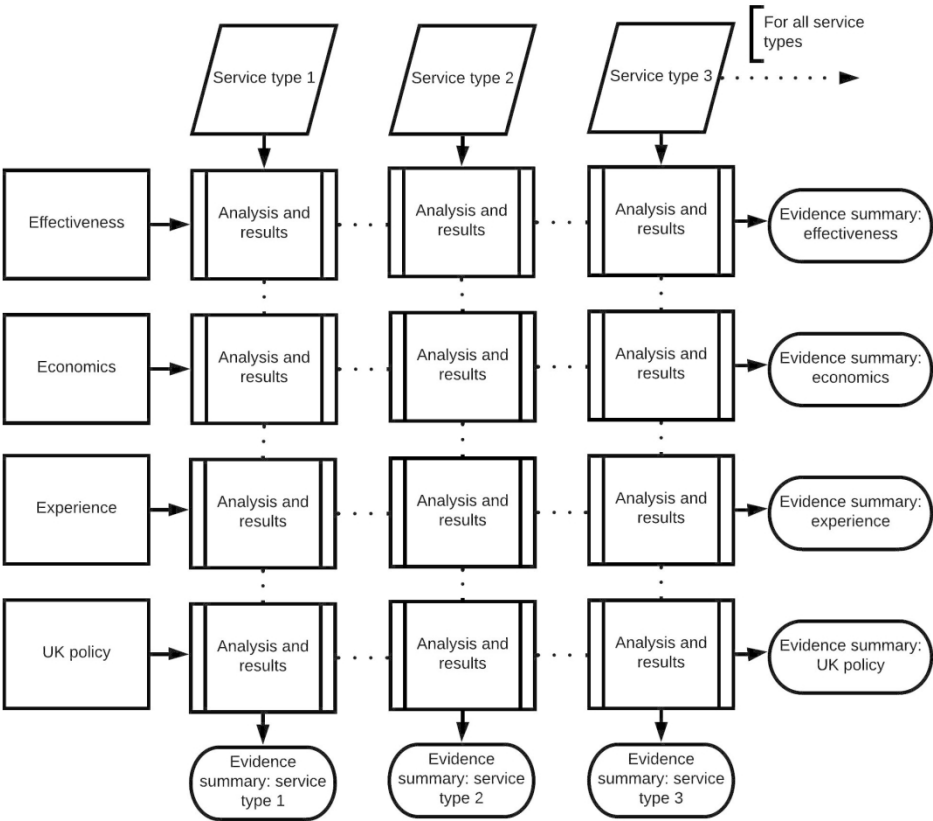
### Data extraction

Development of the conceptual framework



**Overarching synthesis** to finalise a conceptual framework of the programme theory and logic of different service models and to juxtapose intervention effectiveness, costs of care, service users/providers views and policy context.





175x155mm (300 x 300 DPI)

**MEDLINE (Ovid)**

Access via Liverpool University

Database: Ovid MEDLINE(R) ALL &lt;1946 to September 14, 2018&gt;

Search Strategy:

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- 1 exp Respite Care/
- 2 exp Hospice Care/
- 3 exp HOSPICES/
- 4 exp "Hospice and Palliative Care Nursing"/
- 5 exp Day Care, Medical/
- 6 exp Night Care/
- 7 exp Intermediate Care Facilities/
- 8 exp Terminal Care/
- 9 exp HOLIDAYS/
- 10 "day\* away".ti,ab.
- 11 "day care\*".ti,ab.
- 12 "day centre\*".ti,ab.
- 13 "day center\*".ti,ab.
- 14 "day program\*".ti,ab.
- 15 "day service\*".ti,ab.
- 16 holiday\*.ti,ab.
- 17 "home support\*".ti,ab.
- 18 hospice\*.ti,ab.
- 19 "intermediate care".ti,ab.
- 20 "night care\*".ti,ab.
- 21 "night-time care\*".ti,ab.
- 22 "partial hospitalisation\*".ti,ab.
- 23 "partial hospitalization\*".ti,ab.
- 24 "relief care\*".ti,ab.
- 25 "relief support".ti,ab.
- 26 "residential care\*".ti,ab.
- 27 "residential home\*".ti,ab.
- 28 "residential facilit\*".ti,ab.
- 29 respite\*.ti,ab.
- 30 "short break\*".ti,ab.
- 31 "short stay\*".ti,ab.
- 32 "sitting service\*".ti,ab.
- 33 "support program\*".ti,ab.
- 34 "support scheme\*".ti,ab.
- 35 "support service\*".ti,ab.
- 36 "temporary admission\*".ti,ab.
- 37 "temporary break\*".ti,ab.
- 38 "temporary care\*".ti,ab.
- 39 "temporary relief".ti,ab.
- 40 "temporary support\*".ti,ab.
- 41 "short-term admission\*".ti,ab.
- 42 "short-term break\*".ti,ab.
- 43 "short-term care\*".ti,ab.
- 44 "short-term relief".ti,ab.
- 45 "short-term support\*".ti,ab.
- 46 "time off".ti,ab.
- 47 vacation\*.ti,ab.
- 48 "care service\*".ti,ab.
- 49 "overnight stay\*".ti,ab.
- 50 "home-based support\*".ti,ab.
- 51 "befriend\* service\*".ti,ab.
- 52 "short-break foster\*".ti,ab.
- 53 "adult placement scheme\*".ti,ab.
- 54 "shared care".ti,ab.

55 "replacement care".ti,ab.  
56 "family support".ti,ab.  
57 or/1-56  
58 exp Palliative Care/  
59 exp Palliative Medicine/  
60 exp Terminally Ill/  
61 exp Heart Failure/  
62 exp MUSCULAR DYSTROPHY, DUCHENNE/  
63 exp Neoplasms/  
64 exp Muscular Dystrophies/  
65 exp Cerebral Palsy/  
66 exp Spinal Dysraphism/  
67 exp Cystic Fibrosis/  
68 exp Disabled Persons/  
69 exp Disabled Children/  
70 exp Neurodegenerative Diseases/  
71 exp Multiple Trauma/  
72 exp Genetic Diseases, Inborn/  
73 exp Chromosome Disorders/  
74 exp "CONGENITAL, HEREDITARY, AND NEONATAL DISEASES AND  
ABNORMALITIES"/  
75 (advanc\* adj3 disease\*).ti,ab.  
76 (advanc\* adj3 illness\*).ti,ab.  
77 (advanc\* adj3 condition\*).ti,ab.  
78 (advanc\* adj3 disorder\*).ti,ab.  
79 (advanc\* adj3 abnormalit\*).ti,ab.  
80 (advanc\* adj3 impairment\*).ti,ab.  
81 (advanc\* adj3 handicap\*).ti,ab.  
82 (degenerative adj3 disease\*).ti,ab.  
83 (degenerative adj3 illness\*).ti,ab.  
84 (degenerative adj3 condition\*).ti,ab.  
85 (degenerative adj3 disorder\*).ti,ab.  
86 (degenerative adj3 abnormalit\*).ti,ab.  
87 (degenerative adj3 impairment\*).ti,ab.  
88 (degenerative adj3 handicap\*).ti,ab.  
89 (progressive adj3 disease\*).ti,ab.  
90 (progressive adj3 illness\*).ti,ab.  
91 (progressive adj3 condition\*).ti,ab.  
92 (progressive adj3 disorder\*).ti,ab.  
93 (progressive adj3 abnormalit\*).ti,ab.  
94 (progressive adj3 impairment\*).ti,ab.  
95 (progressive adj3 handicap\*).ti,ab.  
96 "diminished life expectancy".ti,ab.  
97 "limited life expectancy".ti,ab.  
98 cancer\*.ti,ab.  
99 duchenne.ti,ab.  
100 dying.ti,ab.  
101 "end of life".ti,ab.  
102 ("end stage renal failure" or "end stage liver failure").ti,ab.  
103 "heart failure".ti,ab.  
104 incurable.ti,ab.  
105 life-limit\*.ti,ab.  
106 "life limit\*".ti,ab.  
107 (life adj3 short\*).ti,ab.  
108 (live\* adj3 short\*).ti,ab.  
109 "life threaten\*".ti,ab.  
110 "limited life expectancy".ti,ab.  
111 LLC.ti,ab.  
112 LLI.ti,ab.

1 113 "muscular dystroph\*".ti,ab.  
2 114 neoplasm\*.ti,ab.  
3 115 "neurodegenerative condition\*".ti,ab.  
4 116 "neurodegenerative disease\*".ti,ab.  
5 117 "neurodegenerative illness\*".ti,ab.  
6 118 "neurodegenerative disorder\*".ti,ab.  
7 119 "neurodegenerative abnormalit\*".ti,ab.  
8 120 "neurodegenerative impairment\*".ti,ab.  
9 121 "neurodegenerative handicap\*".ti,ab.  
10 122 oncology.ti,ab.  
11 123 palliative.ti,ab.  
12 124 "poor prognosis".ti,ab.  
13 125 (serious\* adj3 ill\*).ti,ab.  
14 126 (terminal\* adj3 ill\*).ti,ab.  
15 127 (terminal\* adj3 care\*).ti,ab.  
16 128 (terminal\* adj3 disease\*).ti,ab.  
17 129 (terminal\* adj3 condition\*).ti,ab.  
18 130 (terminal\* adj3 disorder\*).ti,ab.  
19 131 (terminal\* adj3 abnormalit\*).ti,ab.  
20 132 (terminal\* adj3 impairment\*).ti,ab.  
21 133 (terminal\* adj3 handicap\*).ti,ab.  
22 134 (genetic adj3 disease\*).ti,ab.  
23 135 (genetic adj3 disorder\*).ti,ab.  
24 136 (genetic adj3 illness\*).ti,ab.  
25 137 (genetic adj3 condition\*).ti,ab.  
26 138 (genetic adj3 abnormalit\*).ti,ab.  
27 139 (genetic adj3 impairment\*).ti,ab.  
28 140 (genetic adj3 handicap\*).ti,ab.  
29 141 (chromosomal adj3 disease\*).ti,ab.  
30 142 (chromosomal adj3 illness\*).ti,ab.  
31 143 (chromosomal adj3 disorder\*).ti,ab.  
32 144 (chromosomal adj3 condition\*).ti,ab.  
33 145 (chromosomal adj3 abnormalit\*).ti,ab.  
34 146 (chromosomal adj3 impairment\*).ti,ab.  
35 147 (chromosomal adj3 handicap\*).ti,ab.  
36 148 (congenital adj3 disease\*).ti,ab.  
37 149 (congenital adj3 illness\*).ti,ab.  
38 150 (congenital adj3 disorder\*).ti,ab.  
39 151 (congenital adj3 condition\*).ti,ab.  
40 152 (congenital adj3 abnormalit\*).ti,ab.  
41 153 (congenital adj3 impairment\*).ti,ab.  
42 154 (congenital adj3 handicap\*).ti,ab.  
43 155 "complex health\* need\*".ti,ab.  
44 156 "early death\*".ti,ab.  
45 157 "cerebral pals\*".ti,ab.  
46 158 "spina bifida".ti,ab.  
47 159 "cystic fibrosis".ti,ab.  
48 160 encephalopath\*.ti,ab.  
49 161 disabilit\*.ti,ab.  
50 162 disabled.ti,ab.  
51 163 handicap\*.ti,ab.  
52 164 spastic\*.ti,ab.  
53 165 "impaired motor skill\*".ti,ab.  
54 166 "spinal cord condition\*".ti,ab.  
55 167 "multiple trauma".ti,ab.  
56 168 "acquired brain injur\*".ti,ab.  
57 169 "neurological condition\*".ti,ab.  
58 170 "neuromuscular condition\*".ti,ab.  
59 171 "multi-organ disease\*".ti,ab.

172 neurodisabilit\*.ti,ab.  
173 or/58-172  
174 "young adult\*".ti,ab.  
175 "young person".ti,ab.  
176 "young people".ti,ab.  
177 youth\*.ti,ab.  
178 "emerg\* adult\*".ti,ab.  
179 "early adult\*".ti,ab.  
180 (child\* adj3 transition adj3 adult\*).ti,ab.  
181 (adolescen\* adj3 transition adj3 adult\*).ti,ab.  
182 (teenage\* adj3 transition adj3 adult\*).ti,ab.  
183 (paediatric\* adj3 transition adj3 adult\*).ti,ab.  
184 (pediatric\* adj3 transition adj3 adult\*).ti,ab.  
185 "college student\*".ti,ab.  
186 "university student\*".ti,ab.  
187 "post-secondary student\*".ti,ab.  
188 undergraduate\*.ti,ab.  
189 postgraduate\*.ti,ab.  
190 exp Young Adult/  
191 exp ADOLESCENT/  
192 exp ADULT/  
193 exp FAMILY/  
194 exp CAREGIVERS/  
195 exp PARENTS/  
196 famil\*.ti,ab.  
197 carer\*.ti,ab.  
198 caregiver\*.ti,ab.  
199 parent\*.ti,ab.  
200 grandparent\*.ti,ab.  
201 relative\*.ti,ab.  
202 relation\*.ti,ab.  
203 sibling\*.ti,ab.  
204 or/174-203  
205 57 and 173 and 204  
206 limit 205 to yr="2002 -Current"

**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	(Page No.#)
ADMINISTRATIVE INFORMATION			
Title:			
Identification	1a	Identify the report as a protocol of a systematic review	1
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	4
Authors:			
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author	1
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review	17
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	17
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	5
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)	6
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	9
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	10
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	11 and Appendix 1

Study records:			
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	11-12
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)	11-12
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	13-14
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	12
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised	13-14
	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 $\tau$ )	13-14
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)	15
	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)	15
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE)	15

**\* 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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