

# 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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**To cite:** Pilkington G, Knighting K, Bray L, *et al.* 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. *BMJ Open* 2019;**9**:e030470. doi:10.1136/bmjopen-2019-030470

► Prepublication history and additional material for this paper are available online. To view these files, please visit the journal online (<http://dx.doi.org/10.1136/bmjopen-2019-030470>).

Received 19 March 2019

Revised 24 April 2019

Accepted 13 May 2019



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## ABSTRACT

**Introduction** The number of young adults with complex healthcare needs 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 We will comprehensively search multiple electronic databases; use the Citations, Lead authors, Unpublished materials, Google Scholar, Theories, Early examples, and Related projects (CLUSTER) approach, search relevant websites and circulate a 'call for evidence'. Using the setting, perspective, intervention/phenomenon of interest, comparison and evaluation 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 Grading of Recommendations Assessment, Development and Evaluation (GRADE)/GRADE-Confidence in the Evidence from Reviews of Qualitative Research 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

## 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 (CHCNs).
- Aims and review questions were developed with stakeholders from the advisory group including young adults with CHCNs, 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.

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.

## INTRODUCTION

Young adults with life-limiting conditions (LLCs) and young adults with complex physical disabilities often live with multiple comorbidities 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.<sup>1 2</sup> In 2010, there were 55 721 young adults with complex needs living in England<sup>3</sup> and an estimated 100 000 disabled children with complex care

**Table 1** Key definitions

CHCNs	Substantial and ongoing healthcare needs, typically across multiple health concerns, requiring a coordinated response from more than one service
Complex physical disability	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
LLCs	A life-limiting or life-threatening condition where there is no reasonable hope of cure and from which the person is expected to die
Respite care and short breaks	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

CHCNs, complex healthcare needs; LLCs, life-limiting conditions.

needs in England in 2007.<sup>4</sup> 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.<sup>5,6</sup> Table 1 details the 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,<sup>7</sup> improving psychological well-being of parents,<sup>5,8</sup> reducing risk of carer breakdown,<sup>7,9</sup> and avoiding costly unplanned hospital admissions, length of stay or social care intervention.<sup>10,11</sup> 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.<sup>12–15</sup> Seven out of 10 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.<sup>16</sup>

In children services, short breaks provide opportunities for children to enjoy social interaction, support for family carers and support for siblings.<sup>17</sup> Examples include residential schools, sitting services, day care in the home or other settings, or packages tailored to individual needs.<sup>18</sup> 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.<sup>5,9,13,19,20</sup> Limited respite care, particularly for those with very CHCNs, is available for planned short breaks or emergency family situations once young adults with CHCNs have transitioned to adult services.<sup>7,21,22</sup>

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.

## 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.
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 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

A patient and public involvement advisory group (PAG) of young adults and parents has 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.<sup>23 24</sup> 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.<sup>25</sup> 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.<sup>26</sup> The review methods are described in accordance with guidelines (Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Protocols)) for the reporting of protocols of systematic reviews.<sup>27</sup> 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.<sup>28 29</sup> 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<sup>30</sup> 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.

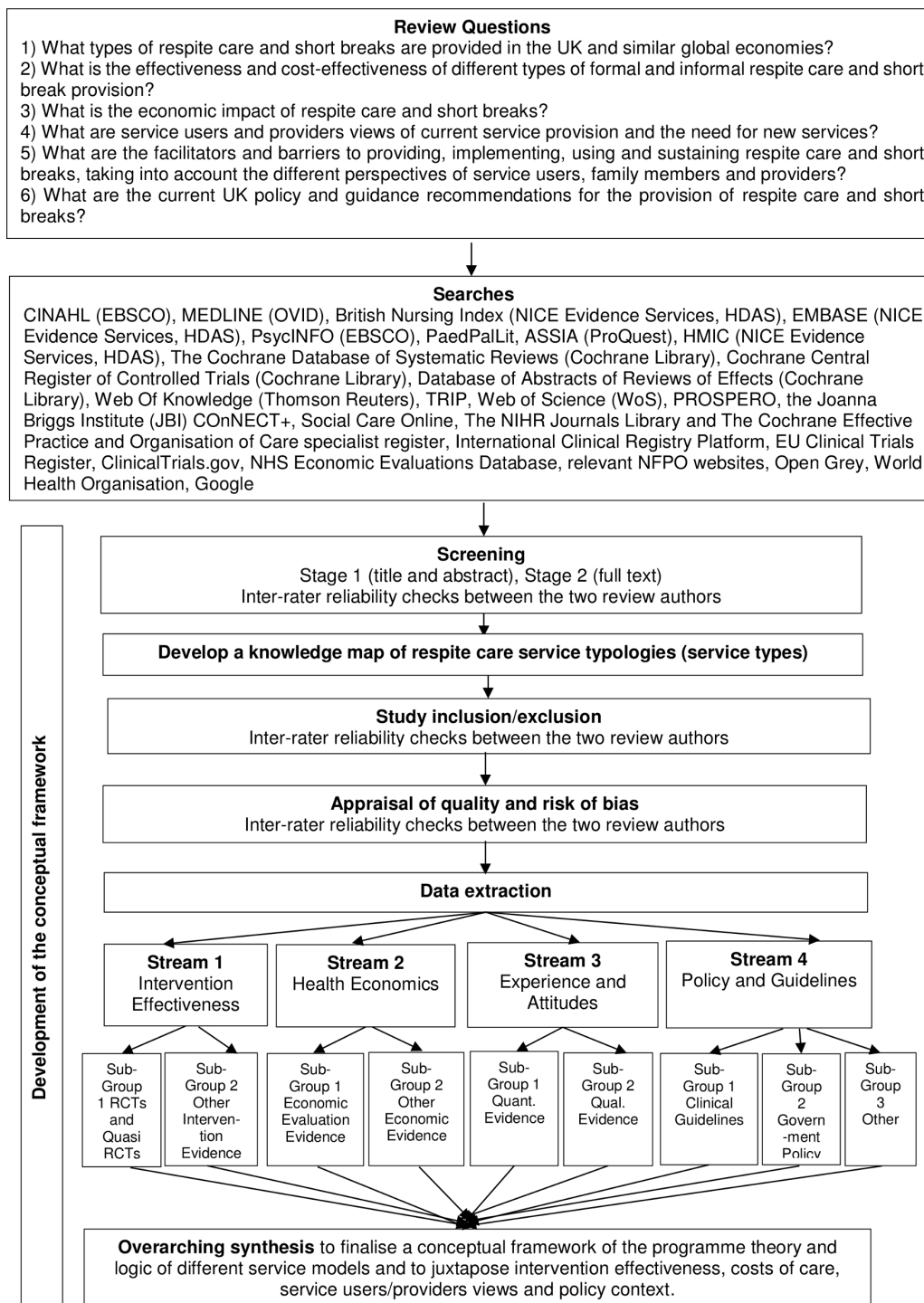
## Information identification

### Search sources

We will search the following electronic databases from 2002 to 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, HMC (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 ClinicalTrials.gov.

We will also search for grey and unpublished literature in Open Grey and Grey Literature Report, and search charity and organisational websites (eg, Together for Short Lives, PaedPalLit and WHO). We will use the Citations, Lead authors, Unpublished materials, Google Scholar, Theories, Early examples, and Related projects (CLUSTER) approach to identify additional outputs (eg, 'sibling' papers or 'kinship' studies) from the included evidence.<sup>31</sup> 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 1st January 2002 due to changes in patient population, service provision and policy change over the last 15 years.<sup>3</sup> 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



**Figure 1** Review processes.

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 keywords, free-text terms and controlled vocabulary (online supplementary 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.<sup>32</sup> Two reviewers will independently screen all titles and abstracts using the inclusion criteria outlined

**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 to current from the 35 OECD countries. <b>Effectiveness:</b> Service user, family, carer and service provider reported quantitative outcomes for example, 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 QALY, cost per admission avoided, other measures for example, 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 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	Outcomes unrelated to effectiveness, experience or economic evidence. Unconfirmed reports and anecdotal opinion for example, newspapers, social media, online blogs. Non-UK policy or guidelines

CHCNs, complex healthcare needs; OECD, Organisation for Economic Cooperation and Development; QALY, quality-adjusted life year.

in [table 2](#). For Stage 1 knowledge map, we will only use the perspective and intervention components of the SPICE criteria for evidence selection, that is, 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 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 PRISMA guidelines, including a flow diagram of included/excluded evidence.<sup>33</sup>

#### 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 checklist as a guide.<sup>34</sup>

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: for example, year, dates and country of data collection, language, source of funding;
- ▶ Methods: for example, study design, duration of follow-up;

**Table 3** Methodological quality assessment tools

Experimental	Randomised controlled trial	Cochrane RoB tool
	Non-randomised controlled trial	Cochrane RoB tool EPOC adaptations for different study designs
	Before and after study	Cochrane RoB tool or National Institutes of Health tool
Observational	Cohort	CASP for cohort studies
	Case-control	CASP for case control studies
	Cross-sectional	CEBMA tool
	Interrupted time-series	Cochrane RoB (EPOC adaptation)
	Case report/case series	CEBMA tool
	Economic evidence	British Medical Journal Checklist for authors and peer reviewers of economic submissions
Qualitative	Qualitative	CASP for qualitative studies
Mixed-methods	Mixed-methods	Mixed-Methods Appraisal Tool
Policy	Policy/guideline document	Appraisal of Guidelines, Research and Evaluation
Other	Grey literature	Appropriate method-specific tool for the type of evidence. If a position statement use the Authority, Accuracy, Coverage, Objectivity, Date, Significance tool to assess the credibility of the source

CASP, Critical Appraisal Skills Programme; CEBMA, Centre for Evidence-Based Management; EPOC, Effective Practice and Organisation of Care; RoB, Risk of Bias.

- ▶ Aims, objectives, hypotheses, target audience;
- ▶ Participant characteristics: for example, 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: for example, 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.<sup>35–45</sup> Disagreements will be arbitrated by a third reviewer until consensus is reached.

#### Data synthesis methods

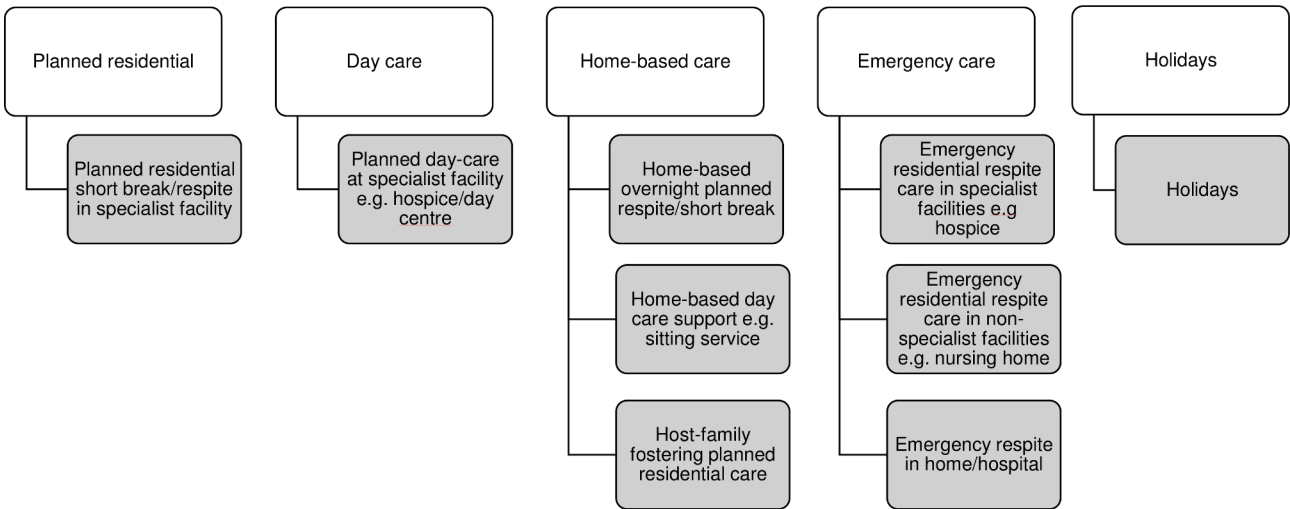
In Stage 1, we will classify the evidence and consider factors such as population, timing and location to create a knowledge map of the different service typologies of respite care. Service typologies will be determined broadly by type, eligibility criteria and target population based on the data extracted 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 (ie, 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 and 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 (eg, 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.<sup>35</sup> Where data are sufficient,

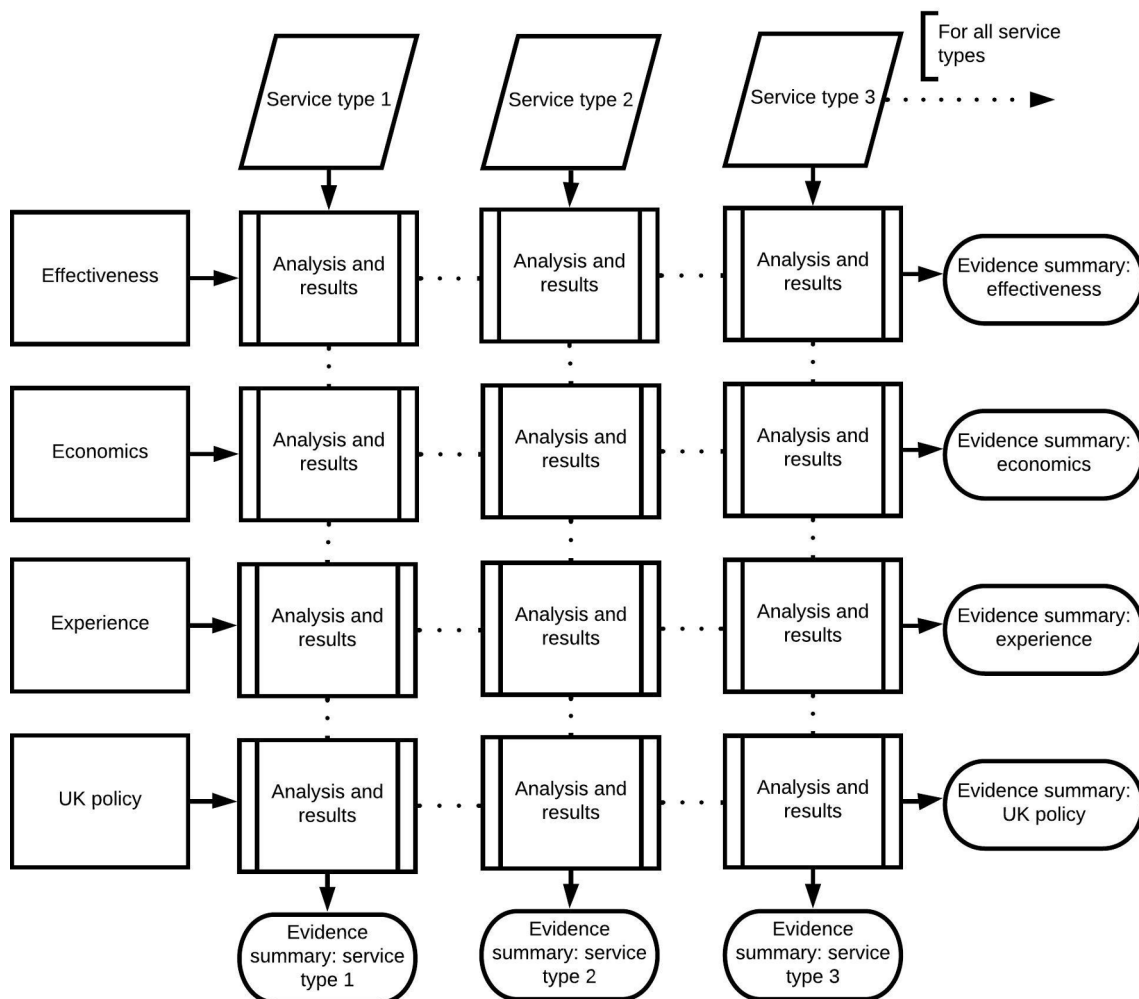


**Figure 2** Initial types of respite care.

we will conduct sensitivity analyses based on missing data and risk of bias criteria (randomisation). Analyses will be conducted using Review Manager (RevMan) V.5.<sup>46</sup>

**Evidence of cost-effectiveness**

We will tabulate and narratively synthesise data derived from economic evaluations (eg, cost-utility and cost-effectiveness, reports of care costs) and other economic evidence (eg, cost of illness or burden of disease studies) by service type. We will consider all direct and indirect



**Figure 3** Evidence matrix.

costs of respite care (eg, direct medical and non-medical costs and productivity losses) including quality adjusted life-year, admission avoided and carer burden, taking account of the following factors: population size, service type, perspective (eg, patient/National Health Service and social services/societal), price year and currency, time horizon, discount rate and type of health-related quality of life instrument, where applicable.

#### Evidence on experience and attitudes

We will include and narratively report the results from qualitative, quantitative and mixed-methods evidence (eg, 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.<sup>47</sup> Findings from the qualitative and qualitative syntheses will be integrated using established methods for combining mixed-methods data.<sup>48</sup>

#### 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<sup>49</sup> to tabulate the evidence based on an a priori framework, following the process outlined for textual analysis.<sup>50</sup>

#### Subgroup analyses

Where possible, we will conduct subgroup 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.<sup>51</sup> We will explore subgroups of interest where data permits, for example, 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 framework method to integrate evidence across the evidence matrix.<sup>52 53</sup> 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.<sup>52</sup> 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)<sup>54 55</sup> 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<sup>56</sup> and examples of good practice<sup>57-59</sup> 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 Stages 1 and 2 findings to ensure that the concepts and relationships are clear and useful for researchers, commissioners, service providers, and policymakers 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 for qualitative evidence.<sup>33 60</sup>

#### 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, BAJ, MM, CM, JN, MRO'B, BR, AT and SS) have contributed to the development of all aspects of the protocol. The review design and methodology were developed by GP, LB, JD, BAJ, MM, CM, JN, MRO'B, 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, BAJ, MM, CM, JN, MRO'B, BR, AT, SS and all authors have critically revised the manuscript and approved the final version and act as guarantors of the review.

**Funding** The review is funded by the National Institute for Health Services Research (NIHR), Health Services & Delivery Research (HS&DR) programme (Study ID 16/115/17). The study is sponsored by Edge Hill University.

**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.

**Patient consent for publication** Not required.

**Provenance and peer review** Not commissioned; peer reviewed for ethical and funding approval prior to submission.

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