Respite care and short breaks for young adults with complex healthcare needs due to a life-limiting condition and/or complex physical disability: Mixed-method systematic review and conceptual framework development

Final protocol July 2018

Funding

This project is funded by the NIHR HS&DR Programme (project number 16/115/17)

Project title

HS&DR Project: 16/115/17 - Respite care and short breaks for young adults with complex healthcare needs due to a life-limiting condition and/or complex physical disability: Mixed-method systematic review and conceptual framework development

Review team:

Dr Katherine Knighting[1], Gerlinde Pilkington[2], Professor Jane Noyes[3], Professor Brenda Roe[1], Michelle Maden[4], Professor Lucy Bray[1], Professor Barbara Jack[1], Professor Mary O'Brien[1], Professor Julia Downing[5], Dr Céu Mateus[6], Professor Sally Spencer[2]

1. Evidence-Based Practice Research Centre, Edge Hill University; 2. Postgraduate Medical Institute, Edge Hill University; 3. School of Social Sciences, Bangor University; 4. Liverpool Reviews and Implementation Group, University of Liverpool; 5. International Children's Palliative Care Network; Edge Hill University; 6. Faculty of Health and Medicine, Lancaster University

Correspondence to:
Dr Katherine Knighting
Evidence-based Practice Research Centre
Edge Hill University
Faculty of Health & Social Care
St Helens Road, Ormskirk
L39 4QP
knightk@edgehill.ac.uk

Table of Contents

List of Figures

List of Tables

1 ABBREVIATIONS AND GLOSSARY

Abbreviations

CCG	Clinical Commissioning Groups
CHCN	Complex healthcare need
CLUSTER	Citations, Lead authors, Unpublished materials, Scholar search, Theories, Early examples, Related projects
GRADE	Grading of Recommendations Assessment, Development and Evaluation
GRADE-CERQual	GRADE-Confidence in the Evidence from Reviews of Qualitative Research
HRQoL	Health-related quality of life
LLC	Life-limiting and life-threatening condition
NFPO	Not-for profit organisation
NHS	National health Service
NICE	National Institute for Health and Care Excellence
NIHR	National Institute for Health Research
PAG	PPI Advisory Group
PPI	Patient and public involvement
QALY	Quality-adjusted life year
RCT	Randomised controlled trial
SPICE	Setting, perspective, intervention/phenomenon of interest, comparison, evaluation
SG	Steering Group
TfSL	Together for Short Lives
TIDieR	Template for Intervention Description and Replication

Glossary

Complex Care	Substantial and ongoing healthcare needs typically requiring a co-ordinated response from more than one sector or organisation. Complex care needs can be the result of chronic illness, disabilities or following hospital treatment. Complex care is sometimes referred to as long-term care or continuing care.
Complex Physical Disability	Complex impairments and/or physical disabilities, often due to congenital or acquired disability, or major neurological trauma, requiring a high level of physical management and support. Sometimes referred to as severe or profound disability. May overlap and interlock with other health conditions or learning disabilities creating a complex patient profile.
Disability	Definition of disability under the Equality Act 2010 is a physical or mental impairment that has a 'substantial' and 'long-term' negative effect on a person’s ability to do normal daily activities.
Formal Respite Care	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	Informal respite care does not involve financial payment for the care provided.
Life-limiting condition	A life-limiting condition is defined as a condition *where there is no reasonable hope of cure and from which the person is expected to die*
Respite Care	Respite care is the temporary provision of formal (paid) or informal (unpaid) physical, emotional, spiritual or social care for a dependent person to promote well-being, independence and to reduce carer distress.
Short Breaks	Together for Short Lives defines short break care as having three main functions: “(1) to provide the child or young person with an opportunity to enjoy social interaction and leisure facilities; (2) to support the family in the care of their child in the home or an alternative community environment such as a children's hospice; and (3) to provide opportunities for siblings to have fun and receive support in their own right”
Young Adults	Typically considered to be 19-25, although some definitions begin at 18 years and extend to 40 or 45. For the purposes of this protocol the definition is 18-40 years of age.

2 PLAIN ENGLISH SUMMARY

Background

People with life-limiting conditions or complex physical disabilities may have complex healthcare needs. The number of young adults with these healthcare needs has risen significantly in the last decade as children with complex conditions survive into adulthood. Respite care and short breaks provide essential support for these young adults and their families, but their needs are often not met after the transition to adult services. Lack of services following transition has a significant impact on both the life expectancy and quality of life for these young adults, including early death, along with increased physical and emotional strain on ageing parents and carers. It is important that the design of future services for this population are informed by the best available evidence, in terms of what works well, for whom, why and in which particular circumstances or settings.

Review objectives

For young adults aged 18-40 with complex healthcare needs, our objectives are to:

- Identify and describe the different types of short breaks and respite care available;
- Assess whether they work and how much they cost;
- Understand the experiences and views of young adults who use short breaks and respite care, their families and carers, and service providers;
- Find out what current guidelines and government policies recommend for respite care services;
- Share what we learn to help develop short breaks and respite care services;
- Make recommendations for new research that will answer gaps found in the evidence.

Review methods

We will look for published and unpublished evidence that investigates respite care and short breaks for young adults with complex healthcare needs. There will lots of types of evidence, for example, academic papers, reports from organisations, and policy documents. In the first instance, we will use this information to create a ‘knowledge map’ which describes the different types of respite care and short breaks currently available, e.g. day services, residential services. Then we will group the different types of evidence within these different types of respite care. The evidence for each type of respite care will then be categorised as one of the following:

1. Effectiveness (whether they work) of respite services for improving care;
2. Comparative costs of respite care;
3. Experiences of respite care including factors that help and those that are barriers to service provision;
4. UK government policy and guideline recommendations for these services.

For each type of respite care, we will extract key information and assess the scientific quality of the evidence to find out what sort of evidence is available for each type of respite care and to identify gaps in the evidence. We will rate our confidence in the strength of evidence for each type of respite care using standard quality grading tools (GRADE and GRADE-CERQual). Then through a process called synthesis, we will then draw together the findings of the evidence in a summary. Throughout the entire process, we will be using the information we gather to develop an overall ‘framework’ of how each type of respite care works, and how different circumstances or settings might change how the respite care works. Once we have completed the review we will work with our steering group and advisory group of young adults and parents/guardians to share the findings using different formats including a final report, papers, conference presentations, and other media such as videos or animations.

3 BACKGROUND AND RATIONALE

This mixed-methods systematic review will focus on young adults with complex healthcare needs (CHCNs) due to life-limiting or life-threatening conditions or complex physical disability. We will use the term life-limiting conditions (LLCs) to indicate life-limiting or life-threatening conditions throughout the protocol.

3.1 Young adults with life-limiting or complex physical disability needs

Whilst there are differences between the populations of young adults with LLCs and young adults with complex physical disabilities, there are many similarities in terms of shared experiences of healthcare services and, increasingly, in the provision or lack of available respite care services to meet their needs. These two populations of young adults are often described as having CHCNs due to a single or multiple diagnosis, for example illness, congenital conditions, or trauma. Many individuals live with multiple co-morbidities and the need for continuous healthcare can be comparable for both populations as there is often an overlap in terms of service needs and provision across a range of conditions and disabilities. These two populations have previously been researched separately. However, with the extended lifespan of these populations there is a growing young adult population who transfer from children's services and require services to meet their CHCNs. There is therefore a clear rationale for exploring the evidence base of both populations together to inform future research and the development of services for young adults with CHCNs. This section provides specific details of how we understand and define the patient population for this systematic review.

3.1.1 Definition of life-limiting conditions

The population of children with LLCs who survive to become young adults is rising annually in England. Due to medical advances, the number of 16-19 year olds with palliative care needs has increased by 45% over the past decade to one in ten young people (1). The latest 2010 data reported 55,721 young adults, aged 18-40, with complex needs living in England (2). The needs of young adults with LLCs are diverse and involve complex life-long symptom, medication management, and palliative care (3). Many children and young people with LLCs die in infancy and childhood, but those surviving into adulthood tend to have degenerative and progressive conditions over many years; resulting in CHCNs requiring daily care provided mainly by family members with support from paid carers, health and social care professionals. The frequency and length of time that care is needed is typically different to adults with terminal illness who frequently require end of life care during the last 12 months of life. The level of care required by young adults is associated with high and increasing costs, which in parallel with the rising population, is placing increasing demands on commissioners and adult providers to meet their needs when they transition to adult services (1,3).

There are over 300 diagnoses, relating to children and young adults with life-limiting and life-threatening conditions originating in childhood which can be grouped into four broad categories (4):

1. Life-limiting conditions where cure is possible but may fail e.g., cancer, irreversible organ failure;

2. Conditions that, though treated intensively over a period of time, inevitably lead to early death e.g., cystic fibrosis;

3. Progressive conditions where treatment is exclusively palliative and often extends over many years e.g., muscular dystrophy;

4. Irreversible, but non-progressive, conditions giving rise to severe disability and sometimes premature death e.g., disabilities following brain or spinal cord insult or severe cerebral palsy.

Drawing on key terms from the literature and the definition from Together for Short Lives (TfSL), the UK charity for children, young people and young adults who are expected to have short lives, we have defined a young adult with LLCs as follows:

> Young adults with a life-limiting or life-threatening condition, where there is no reasonable hope of cure and from which they are expected to die.

3.1.2 Definition of complex physical disability

Over the last 13 years, the prevalence of children and young people with severe disability and complex needs has risen due to the increasing survival of babies and children who have experienced severe trauma or illness (4,5). In 2007, it was estimated that there were 100,000 disabled children with complex care needs in England (6) and this number is estimated to have risen by 50% in the last decade (4). There is an urgent need to gather evidence on the life experiences of the rising number of young adults with complex healthcare conditions, to explore the needs of this patient population and to assess their implications for future demand of services (4).

There is wide variation in the definitions of disability and levels of severity (5). There is also variation between the definitions of disability for the children and young people compared with the adult population. The Equality Act 2010 defines ‘disability’ as a physical or mental impairment that has a 'substantial' and 'long-term' negative effect on a person’s ability to do normal daily activities (6). Complex physical disability can be grouped into three broad categories (7):

1. Sudden onset conditions, e.g. acquired brain injury, spinal cord conditions, peripheral nervous system conditions, multiple trauma;

2. Progressive and intermittent conditions, e.g. neurological and neuromuscular conditions, severe musculoskeletal or multi-organ disease, or physical illness/injury;

3. Stable conditions (with/without degenerative change) e.g. congenital conditions, post-polio or other previous neurological injury.

Complex physical disability is sometimes referred to as ‘severe’ or ‘profound’ disability and may overlap with other health conditions creating a complex patient profile. The team acknowledges that complex patient profiles can often include learning disability or cognitive impairment, however the focus of this review is on healthcare needs so eligible studies will be limited to young adults with a CHCN due to complex physical disability. Given the variable definitions of disability for children and adults, we will also include young adults with a complex physical disability due to a cancer diagnosis which originated in young adulthood to match the inclusion of cancer in the children’s definition of complex physical disability.

For this review we have defined a young adult with complex physical disability as follows:

Young adults with impairments and/or physical disabilities, due to congenital or acquired physical disability, or major neurological trauma, which require a complex level of physical management and support.

3.1.3 Definition of complex healthcare needs

Defining the concept of ‘complex’ is challenging as it can be used to describe or indicate many things depending on the setting and perspective (8). The healthcare needs of this young adult population may range from complex to highly complex. For example, young adults who are dependent on long term ventilation or have complex drug regimens are often considered too complex for many respite care services, resulting in them being unable to access universal respite care and requiring specially commissioned services. The variation in terminology, the spectrum of complexity, and inflexibility of adult assessment processes can result in inequality of care and loss of funding for services, including respite care. Therefore, using a broad definition will encompass all relevant evidence. Whilst there is no standard agreed definition of CHCNs (3); it is typically used to refer to physical, mental and/or health needs, which can affect people in different and often multidimensional ways. It has been argued that the term ‘complex’ relates more to the complexity of service provision rather than individual needs and that the term ‘multifaceted condition’ may better describe the interconnectedness of an individual’s varied health and social care needs (9). However, CHCNs is a common term in the literature. Common elements across the definitions in the literature suggest that complex needs can be considered both in terms of breadth (wide range of needs) and depth (high level of needs) (10). We have defined complex healthcare needs as follows:

> CHCNs are substantial and ongoing healthcare needs typically across multiple health concerns and requiring a co-ordinated response from more than one service.

3.1.4 Definition of young adult

There is no universal consensus on the definition of a young adult in the UK, e.g. the Ministry of Justice uses the age-band 18-20 years, the National Health Survey for England uses 16-24, and the British Crime Survey uses 18-25 years (11). Services in the UK provide care for different age groups and social services tend not to define respite services by age group, so it is important to use a sufficiently broad age range. Depending on the service and location, services for children with CHCNs may be extended beyond 18 years of age to an individually-defined upper limit to provide services for young adults, for example, up to 23 years at Claire House Children’s Hospice (https://www.clairehouse.org.uk/), 35 years at St Elizabeth Hospice (https://www.stelizabethhospice.org.uk/how-we-can-help/hospice-care/young-adult-service.aspx), or 40 years at The J’s Hospice (https://www.havenshospices.org.uk/thejs), and many adult National Health Service (NHS) services begin at 16 or 18 years. It is recommended that transition plans from children’s to adult services begin when the child is 13-14 years, although due to individual preferences this is likely to vary (12,13). Drawing on key definitions from the literature (2,14), feedback from stakeholders, currently available services in the UK, and TfSL, we have defined young adults as follows:

> Young adults are defined as people aged 18-40 years.

3.2 Respite care and short breaks for young adults with complex healthcare needs

Respite care and short breaks are an essential component of ongoing support for children, young people, and young adults with CHCNs (15,16). They provide a break from caring, with multidimensional benefits for all members of the family (17,18). Together for Short Lives (TfSL) defines short break care as having three main functions: "1) to provide the child or young person with an opportunity to enjoy social interaction and leisure facilities; 2) to support the family in the care of their child in the home or an alternative community environment such as a children's hospice; and 3) to provide opportunities for siblings to have fun and receive support in their own right" (18). Typically, such provision includes residential hospice care or similar service, day care, host-family respite, and home-based support including sitting services and holiday cover. Respite care and short breaks are provided by both formal and informal carers; formal carers are typically defined as registered professionals or care staff who either work privately or for provider organisations, or receive payment for their services. Informal carers are often family members or friends who provide the same type of care on an unpaid basis, though some informal carers may receive payments through personal care budgets managed by families. This section provides specific details of the current provision of respite care and short breaks, including elements which shape the potential outcomes for this systematic review, and our definition of the intervention.

3.2.1 Current service provision

There are clear differences between child and adult services in the way that respite care is conceptualised, funded and provided (19). Typically the term 'short breaks' is used in children's services to encompass all levels of care, whether residential or in the home, and is a key service provided by children's hospices, and some specialist children's services (20). Planned respite care in adult services focuses on the need to give the carer a break from caring rather than providing opportunities for the person receiving care, and is typically referred to as 'respite' or 'replacement' care. Respite care and short breaks for young adults may be provided by child-oriented services which are unsuitable for young adults, and there is variation across providers and commissioners in what is considered as the upper age-limit for children's services. Typical adult services predominantly serve the needs of older people, those with a cancer or other terminal diagnosis, and people requiring end of life care, rather than fluctuating health conditions and may be inappropriate for respite care for young people with CHCNs due to a LLC or complex physical disability (15,21–24). With notable exceptions such as cystic fibrosis and long term ventilation, adult sector staff within the UK generally have little experience of paediatric conditions in adults, or supporting young adults with complex needs (3,12,23,25). Limited respite care, particularly for those with very complex health needs, is available for planned short breaks or emergency family situations once young adults with CHCNs have transitioned to adult services (3,12,26).

There is considerable variation in the definition of ‘residential short breaks’ for young people with disabilities used by social care authorities, ranging from residential schools, sitting services, and day care in the home or other settings, to flexible packages tailored to suit individuals (5). This mirrors other studies of local authority definitions which have identified a myriad of different ways in which the services may be named, commissioned, paid for and delivered, even within the same authority (27). However, estimates from local authorities suggest that only eight in every 10,000 disabled children aged 0-17 years receiving social care services, and 18% of children receiving a service from disabled children’s teams, were receiving residential short breaks (5).

The nature and costs of respite care may vary considerably depending on the provider and level of complex health needs to be supported and estimating costs may therefore be a complex process. Referral, assessment models and procedures may also vary between services and the care required by young adults with CHCNs is highly individual. Following transition to adult services, there are legal and policy changes to care provision including the transfer of parental to personal responsibility, unless there are capacity issues, that impact on decision making and care planning. Many families are ill-prepared for these changes. This can include significant changes in other areas of life, for example where the focus of any assessments for support with housing or welfare support moves from being the whole ‘family’ (such as using parental income and other dependents to assess need) to assessment of the young adult alone, with their family largely disregarded in the assessment process. Consequently, young adults can face significant barriers to accessing the right care and support as they make the transition to adult services (28,29). Parents have described the transition process as ‘like falling off a cliff’ when the support from children’s services ends and appropriate adult services are not in place, adding to the complex burden of living with CHCNs for young adults and their families (30).

3.2.2 Benefits of respite care and short breaks

Current evidence indicates that respite care and short breaks have a broad range of benefits such as increasing family carer resilience (26), improving the psychological well-being of parents (15,31), reducing the risk of carer breakdown (22,26), and avoiding costly unplanned hospital admissions, length of stay or social care intervention (32,33).

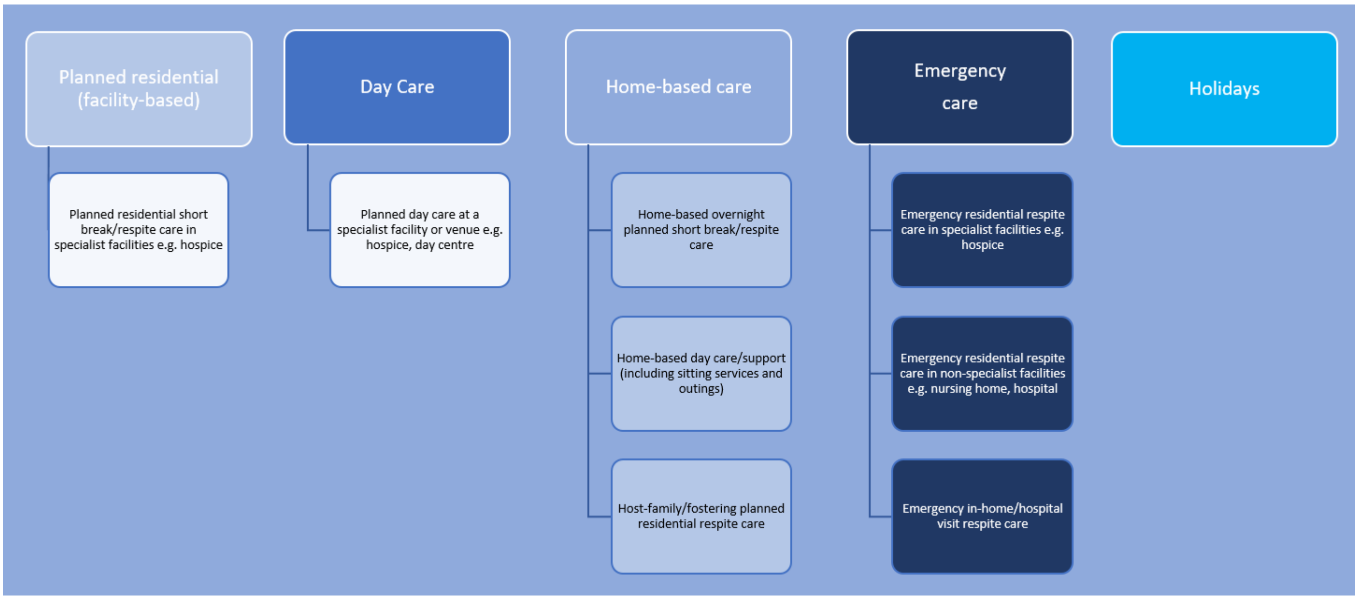
Much of the available evidence on the use and impact of respite care and short breaks relates to children's services, such as hospices, rather than services for young adults with LLCs. This is because until relatively recent improvements in medical care, few of them survived into adulthood. However, more people are now surviving beyond childhood where the needs of young adults with LLCs may in fact increase as they mature, when they may wish to live independently, and their parents develop their own health problems as they grow older. With a rapidly growing population of young adults making the transition from child to adult services there is increasing evidence of poor continuity of care, including respite care provision, resulting in the young adult and their family having unmet needs; adversely affected social, educational, vocational and spiritual outcomes; inadequate management of complex co-morbidities; deterioration in the young adult's physical and mental health; family carer burnout; and inappropriate, costly hospital admissions (23,34,35). Most disturbingly, earlier death may result from poor transition and loss of services (35).

3.2.3 Definition of respite care and short breaks

A systematic review of respite care provision for older people with dementia identified eight models of respite care and short breaks, and characterised services according to: duration, pattern of use, location, response (e.g. planned or emergency care), and the characteristics of service users and staff (36). The models of respite care included: day care; home day care; clubs, interests or activity groups; home-based support; host-family respite; overnight respite in specialist facilities; overnight respite in non-specialist facilities; and holidays (36). Other models such as emergency residential respite and emergency home based respite are also described in the literature. These models reflect many of the known service types for young people with LLCs and complex physical disability, demonstrating the variation in services configuration; it is also likely that other models will evolve in response to growing demand.

From an initial scoping of the evidence to inform the protocol and previous work of the team we have identified an initial list of nine service types depicted in Figure 1 grouped into five overarching sets determined by response and location for ease of presentation. It is also acknowledged that some providers will offer more than one type of service.

Figure 1. Initial service types of respite care grouped by purpose and location



The definitions of short break and respite care used by children and adult services differ by service type and intended outcomes. The provision of care requires the flexibility to meet the needs of the individuals needing care and those providing care. Some of the factors that influence service delivery may include (33):

- Location (e.g. in the person's own home, at a carer's home, residential or community setting)
- Duration (e.g. for a few hours, overnight, several days)
- Timing (e.g. weekdays, weekends, evenings)
- Provider (e.g. local authorities, health agencies, voluntary/independent agencies)
- Payment for care (e.g. use of personal budget, care package, provider or charity funded)

Drawing on the literature and policy statements we will use the following definitions for respite care and short breaks:

> Respite care and short breaks are the temporary provision of formal (paid) or informal (unpaid) physical, emotional, spiritual or social care for a dependent person.
>
> 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.

3.3 Importance of the review

Children and young people with CHCNs, and therefore young adults, have multiple co-morbidities and/or disabilities, so in addition to their primary diagnosis or condition, they are often susceptible to other conditions. Care for these young people and adults is an ongoing, complex process, with no simple care pathway, and often multiple, unplanned episodes of illness. ‘Aiming High for Disabled Children: Better support for families’ (37) made a clear policy commitment to improving available data on disabled young people and their access to services but there is much left to achieve including improving access to specialist services such as short breaks/respite care (38). Seven out of ten families caring for someone with profound or multiple disabilities report having reached, or come close to, ‘breaking point’ due to a lack of short break services (39).

The Care Quality Commission (CQC) found a significant shortfall between policy and practice during transition from child to adult services due to fragmentation of the system, which can be confusing and difficult to navigate for young adults with CHCNs, their families and staff caring for them (12). This is supported by evidence showing that poor service provision following transition to adult services has a significant impact on both the life expectancy and quality of life for these young adults, including early death, along with increased psychosocial burden on families and carers (19,23,34,35). Previously published research by the review team (35,40) and a national survey of hospices and healthcare professionals conducted by the team in 2015 identified significant gaps and challenges in providing respite care for young adults with CHCNs and the need for robust evidence to inform service development (41).

Commissioners and service providers have a statutory duty under the Children & Families Act 2014 (42) and the Care Act 2014 (43) to ensure seamless provision of responsive, appropriately funded and integrated services for young adults with CHCNs as they transition to adult services (1,12,21). Despite the rising number of young people with CHCNs surviving into early adulthood and the consequent escalation in care service demand for themselves and their families, the current scale, cost and types of available respite care have not been collated and evaluated at a national level. Comprehensive data collation is challenging due to the range of public and private providers, fragmented development of independent services and the different funding methods available, including commissioned care (NHS or social care), local authority, charity funded, and use of personal budgets.

Evidence on the effectiveness and cost-effectiveness of respite care/short breaks, and the views and experiences of service users, is published in a variety of sources across the evidence spectrum. Given the uncertainties concerning types of available care and lack of clarity on the optimum models of service provision, it is essential to systematically review the plethora and diversity of available evidence, and to integrate it in a cohesive summary, highlighting gaps in the evidence-base to inform future research.

4 AIMS AND OBJECTIVES

The aim of this mixed methods review is to identify, appraise and synthesise evidence relating to the type and impact of respite care and short breaks provision for young adults (18-40 years) with CHCNs; to explore service intention, service user perspective, factors that may limit or facilitate the delivery of such care, policy intentions and cost-effectiveness in order to develop a conceptual framework for respite care and form the basis of recommendations for future service development and the need for new research.

To achieve the above aim, our objectives are:

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.

4.1 Systematic review questions

For young adults (18-40 years) with CHCNs due to a LLC or complex physical disability:

1) What types of respite care and short breaks are provided in the UK and similar global economies? (Objective 1)

2) What is the effectiveness and cost-effectiveness of different types of formal and informal respite care and short break provision? (Objective 2)

3) What is the economic impact of respite care and short breaks? (Objective 2)

4) What are service users’ and providers’ views of current service provision and the need for new services? (Objective 3)

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? (Objective 3)

6) What are the current UK policy and guidance recommendations for the provision of respite care and short breaks? (Objective 4)

5 METHODS

The team acknowledges that execution of the protocol may change during the systematic review process due to the complexity of the mixed-methods approach and the nature of the evidence. Deviations from the planned protocol will be recorded in the final report.

5.1 Summary

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 will be undertaken to create a third, integrated synthesis (44). The review will be undertaken to evaluate the effectiveness of respite care and short breaks for improving outcomes; to identify and evaluate the comparative costs of respite care and short breaks; to explore stakeholder experiences and attitudes, and the facilitators and barriers to implementation and sustainability of respite care and short breaks; and to collate and evaluate relevant UK policy and guidelines to contextualise the relevant evidence for the UK. Throughout the process, we will build a conceptual framework to articulate the programme theory (i.e. how the service types are intended to work, what they aim to achieve, what outcomes they have and for whom) and the programme logic (i.e. core elements and processes to achieve the outcomes) for the different respite care service types identified.

We will use a similar two-stage approach to that used in other mixed-methods systematic reviews (45,46) to achieve the review objectives set out in Section 4. The two-stage approach allows for the systematic review to address broader questions, consider evidence relating to less well-defined interventions, to promote dialogue with stakeholders, and to ultimately ensure the review is relevant for end-users (45). See Figure 2 for details.

Stage 1 – Developing a knowledge map of respite care service typologies

We will screen the searches using explicit inclusion and exclusion criteria to identify the evidence. We will then undertake a mapping exercise to identify, organise, and describe different service typologies 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.

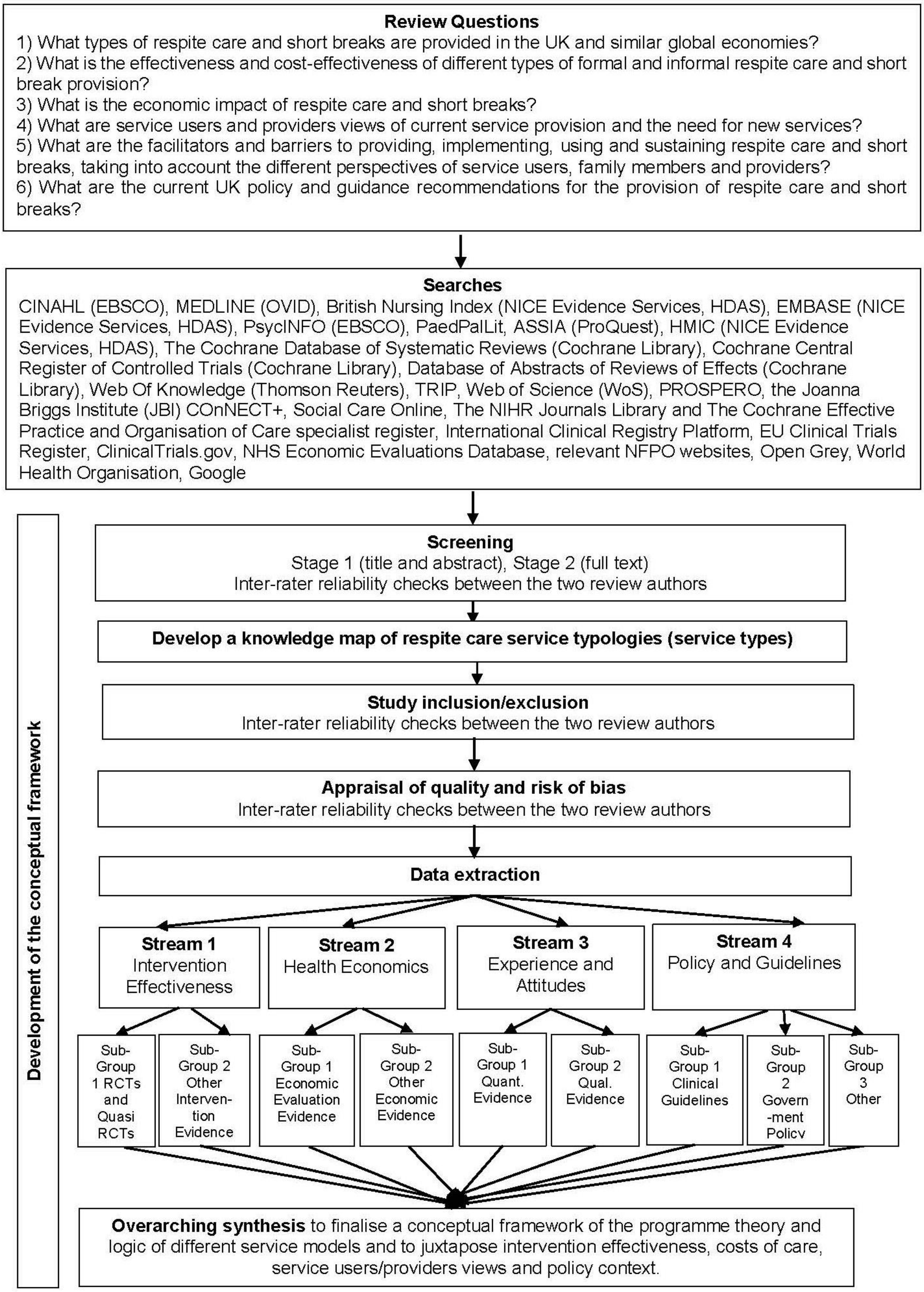
Stage 2 – Evidence review

We will categorise and group the evidence by the service typologies identified in Stage 1 and by four types of evidence (intervention, qualitative, economic, policy) to form an evidence matrix. We will extract key descriptive information about each item of evidence, evaluate its quality and extract key results/recommendations. The findings will be integrated in an overarching evidence synthesis, which will be used alongside the knowledge map of respite care services to develop a conceptual framework of the essential elements of service provision as described above.

Developing a conceptual framework

Building on the service typologies identified in Stage 1, we will develop a conceptual framework throughout Stage 2 based on the overarching synthesis and stakeholder involvement. We will develop the programme logic and a series of logic models for each typology as described above to show the different designs, inputs, processes and intended outcomes for various stakeholders (47).

Figure 2. Mixed-method systematic review flowchart



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

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.

5.2 Identifying the literature

The SPICE (Setting, Perspective, Intervention/phenomenon of Interest, Comparison, Evaluation) framework (48) has been used to inform the search strategy and underpins the approach to Stages 1 and 2.

Table 1. SPICE inclusion and exclusion criteria

	Inclusion	**Exclusion**
Setting	Services and providers of formal respite care and short breaks including hospices, residential care homes, adult day services, individual providers and paid carers/family carers working within young adults' home settings, and informal care from unpaid family members.	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 (Section 3.5) 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 who do not require respite care/short breaks.
Intervention/ phenomenon of interest	Formal (paid) and informal (unpaid) respite care/short breaks (Section 3.2.3) in relation to intervention effectiveness, cost-effectiveness, stakeholder experience and attitudes, UK policy and guidance.	Care other than respite care and short breaks.
Comparison	Any type of formal and 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 will be included. **Intervention effectiveness:** Service user, family, carer, and service provider outcomes such as: quality of life, well-being, health impact, stress and coping, family cohesion or satisfaction with care. We will also include any other quantitative measures reported. **Cost-effectiveness:** Information on UK costs: evaluations of the economic impact of respite care such as incremental cost per quality-adjusted life year (QALY) or cost per admission avoided. We will also collect other measures associated with the costs of providing respite care such as staff grade, time, equipment and transport, to estimate relevant and relative costs for each type of care provision. **Experience and attitudes:** Concepts and themes emerging from recognised qualitative methods (e.g. grounded theory analysis, thematic analysis, framework analysis) that capture attitudes, beliefs, preferences and opinions on the provision of respite care, along with all other potential outcomes. **Policy and guidelines:** Recommendations, directives or actions and anticipated outcomes identified in UK policy statements or guidelines.	**Streams 1 and 2** Outcomes unrelated to effectiveness, experience, or economic evidence. **Stream 3** (experience and attitudes) unconfirmed reports and anecdotal opinion e.g. newspapers, social media, online blogs. **Stream 4** Non-UK policy or guidelines.

5.2.1 Search sources

The search strategy will be developed by the wider review team in conjunction with an experienced information specialist to identify relevant published and unpublished evidence (e.g. primary studies, evaluations, policy documents) to answer the review questions (see Section 4.1). The search strategy will initially be developed for MEDLINE using a combination of controlled vocabulary, for example Medical Subject Headings (MeSH) and free-text terms which will be translated for use in other resources. A sample search strategy is provided in Appendix 1. The search strategy will be underpinned by the inclusion criteria, which have been developed using the SPICE framework to reflect the complexity of the evidence base (48).

A comprehensive overall strategy will be used to search the following electronic databases from 2002-onwards:

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), Database of Abstracts of Reviews of Effects (Cochrane Library), Cochrane Effective Practice and Organisation of Care Specialist Register, Web Of Knowledge (Thomson Reuters), TRIP (https://www.tripdatabase.com/), Web of Science, PROSPERO (https://www.crd.york.ac.uk/PROSPERO/), the Joanna Briggs Institute COnNECT+ (http://connect.jbiconnectplus.org/), Social Care Online (http://www.scie-socialcareonline.org.uk/), and the NIHR Journals Library (https://www.journalslibrary.nihr.ac.uk/#/).

To further identify evidence for each specific stream, the strategy will be adapted and applied to the following databases:

- For **Stream 1** (intervention effectiveness) we will also search trials registers including the Cochrane Central Register of Controlled Trials (Cochrane Library), International Clinical Trials Registry Platform (http://apps.who.int/trialsearch/), EU Clinical Trials Register (https://www.clinicaltrialsregister.eu/ctr-search/search) and ClinicalTrials.gov (https://clinicaltrials.gov/) to identify ongoing and recently completed trials.
- For **Stream 2** (health economics), we will also search the NHS Economic Evaluations Database (Cochrane Library).
- For **Stream 4** (policy and guidelines) evidence will also be identified through internet searches (Google, Google Scholar), relevant NFPO websites, hand searching, and consultation with the Steering Group (SG) and PPI Advisory Group (PAG).

5.2.2 Searching for grey and unpublished literature

Results from scoping searches suggest that relevant information is likely to be found within the grey literature, for example, central and local government evaluations and impact assessments of personal budget pilots and 'satisfaction' surveys for specific services published in reports or annual reports, or unpublished data produced by third-sector organisations. To inform both stages of the review we will conduct a broad search for grey and unpublished literature via Open Grey (formerly SIGLE - System for Information on Grey Literature in Europe http://www.opengrey.eu/), Grey Literature Report (http://www.greylit.org), World Health Organisation (http://www.who.int/en/) and Google. We will attempt to identify evidence from as many sources as possible, including:

- Asking SG and PAG members to identify relevant known literature
- Asking SG and PAG members to identify topic experts, useful websites, and organisations to contact
- Scanning relevant websites for relevant literature
- Targeting topic experts, stakeholders, and service providers through a 'call for evidence' which will aim to identify completed or ongoing reports or evaluations. The call for evidence will be shared through networks, direct emails, and using social media.

A list of potentially relevant websites and organisations to search and/or approach for information is presented in Appendix 2. Results from the above methods of searching will be tabulated to record source data (organisation/expert/URLs), date contacted/found, and the references identified through that method.

In addition to examining the reference lists of included evidence identified through database searching, a purposive and iterative approach to searching the literature will be undertaken following the CLUSTER approach (49). The CLUSTER approach aims to identify additional relevant outputs that may include a 'sibling' paper (i.e. papers from the same study – for example, qualitative studies, economic evaluations or process evaluations associated with an RCT) or 'kinship' studies that inform relevant theoretical or contextual elements. **Error! Reference source not found.** shows the key details of this approach which emphasises the need to adopt multiple search techniques (e.g. citation searching, 'key pearl' searching, ancestral searching) to identify relevant grey literature and associated relevant studies. It aims to identify additional material associated with a study of interest, rather than those simply using the same terminology, thus overcoming one of the limitations of database searching. We will test and refine the strategy using an iterative process to ensure that all known potential studies are included and non-relevant studies are excluded.

Table 2. The CLUSTER approach

Element	**Search procedure**	**Sources**
Citations	Identify at least one 'key pearl' through consensus with review team	Preliminary searches of databases and grey literature
Lead authors	Check reference list of 'key pearl', conduct lead author search	Full text of 'key pearl', search of reference management collection, Google (e.g. institutional repository, author publication webpage)
Unpublished materials	Make contact with lead author	Email
Scholar searches	Citation searches on 'key pearl' and other relevant studies. Conduct search of 'project name'	Web of Science/Google Scholar
Theories	Follow up 'key pearl' and other cluster documents for citations of theory. Recheck for mention of theory in titles/abstracts/keywords, iterative searches for theory in combination with condition of interest	Full text of 'key pearl', search of reference management collection, databases
Early examples	Follow up key pearl citation and other cluster documents for citations to project antecedents and related projects	Full text of 'key pearl'
Related projects	Conduct named project and citation searches for relevant projects identified from cluster documents, seek cross case comparisons by combining project name/identifier for cluster with project name/identifiers for other relevant projects	Web of Science/Google Scholar, databases

Where possible, we will initiate search alerts in the source databases to identify additional relevant studies as the review progresses. The final date for evidence to be included in the review will be 28th February 2019. Bibliographic details of evidence identified after this date will be included in the appendices of the review. Results from the searches of multiple electronic databases and other sources will be combined and de-duplicated in a single file to maximise search efficiency.

5.3 Stage 1 - Developing a knowledge map of respite care service typologies

One of the main objectives of this systematic review is to identify and characterise the different service typologies 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. The knowledge map will draw upon a broad range of evidence in order to identify service types and specifications of respite care and short break care, and will inform a gap analysis.

De-duplicated results of the searches of electronic databases will be uploaded to Covidence, a web-based software platform used to store information and manage each stage of the systematic review (50). Two reviewers will independently screen all titles and abstracts to identify potentially relevant evidence, then full-text versions of potentially relevant evidence will be assessed for inclusion using the criteria outlined in **Error! Reference source not found.**. Where evidence from a study is reported across multiple publications, it will be coded using a single core reference. Disagreements will be resolved through discussion and consultation with a third reviewer where necessary. The bibliographic details of evidence that was excluded because it did not meet our inclusion criteria will be available on request as an electronic addendum to the final report.

This mixed-methods systematic review will focus only on populations that can be described as having CHCNs; however, many respite services provided by organisations and local authorities are commissioned for people with a range of different needs. Some of the evidence we find is likely to have a mix of those who match the population of interest and those that do not. It is important to note that the focus for this research is complex healthcare needs regardless of where the respite care is provided, and as such, populations with solely educational or social care needs will be excluded. Studies will be included where young adults are part of a larger sample, when it is possible to identify data from young adults separately to children and young people under the age of 18, or adults over the age of 40 years. It is recognised by the team that some flexibility may be needed when assessing the relevance of the available evidence within studies and additional information may be sought from authors or services to clarify relevance.

Table 3. Inclusion criteria for the knowledge map

Inclusion	Exclusion
Any evidence (study, evaluation, example of service model) about respite care or short breaks in any setting for young adults (*18-40 years) with LLCs and/or complex physical disability *Including those where no upper age limit is stated.	Care other than respite care and short breaks Services specifically commissioned for young adults with learning disability or mental health needs *Evidence which describes young adults below the age of 18 or above 40 years of age. Young adults who do not require respite care/short breaks.

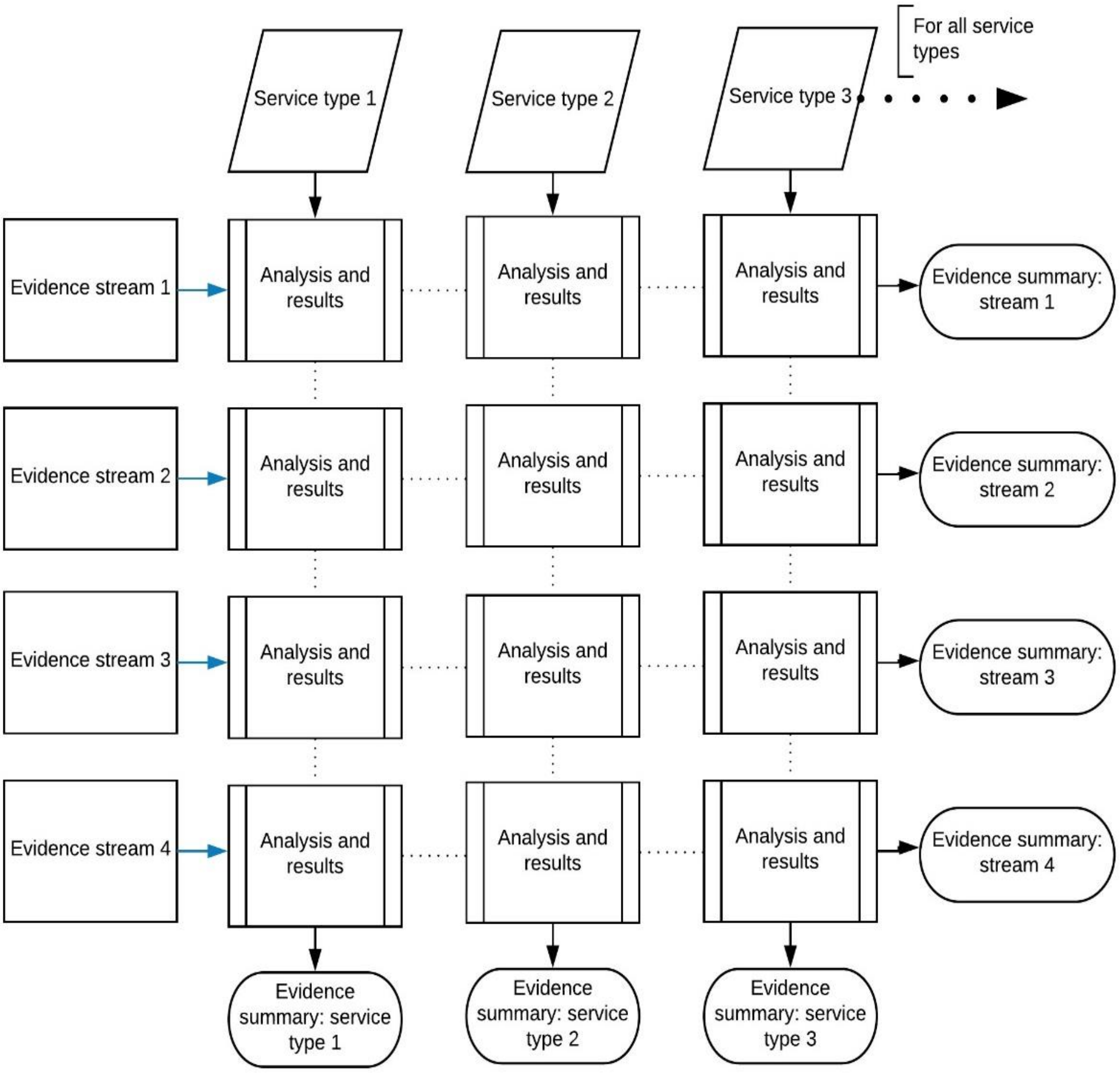
We will extract bibliographic details alongside detailed information relating to the intervention description using the Template for Intervention Description and Replication (TIDieR) checklist as a guide (51), including data relating to programme theory, logic, and design. The team will develop and apply keywords to describe and categorise the types of respite care reported. Using the service types described in the background section (3.2) as an initial starting point, the included evidence will be used to create a knowledge map of the different service typologies of respite care, taking into account the population, timing, location, and level of care provision. We will provide a narrative summary of the service typologies identified, and the results will be discussed with the PAG and SG to ensure fitness for purpose. The bibliographic details of evidence included in the knowledge map will be available on request as an electronic addendum to the final report.

5.4 Stage 2 – Evidence review

Evidence included in Stage 1 will then be considered for inclusion in Stage 2 using the full SPICE criteria outlined above in **Error! Reference source not found.**1 (5.2). Due to the anticipated uneven distribution of the evidence, the team may need to implement a sampling frame to ensure that there is a representative sample of conditions. Bibliographic details of excluded studies will be tabulated with reasons for exclusion.

The 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 studies (52). The team will build on the service typologies of respite care identified in Stage 1 and categorise the evidence by service type and evidence stream using a matrix approach (see Figure 2). Evidence will be examined by a) service type, to provide summaries of the breadth and depth of evidence for each type of service and, b) by evidence stream to provide clear information on the strengths and limitations of the evidence base. This will help to characterise the strength of evidence that supports each type of service, to inform future service development for commissioners and service providers, and for future research. Further information about how the matrix will be used for the overall synthesis is presented below. Throughout Stage 2 the team will develop a conceptual framework with a programme theory and programme logic for each service type.

Figure 3. Conceptual and methodological matrix for synthesis by service type and evidence stream



Where feasible, we will include non-English language studies in Streams 1-3; however, only UK-specific evidence, written in English language will be included for Stream 4 and for grey literature. To address the review questions outlined in section 4.1, we are expecting to include the following types of evidence:

Evidence Stream 1 Intervention effectiveness (review questions 1-2)

Quantitative evidence of the effectiveness of the intervention (respite care and short breaks): randomised, quasi-randomised controlled trials or other intervention studies, such as before and after studies or observational cohort studies, evaluating the effectiveness and outcomes of care. We will include any control or comparison group, for example respite care versus no respite care or hospice short breaks versus home-based short breaks, and studies with no comparison group.

Evidence Stream 2 Health economics (review questions 1-3)

Quantitative evidence relating to health economics: economic evaluations e.g. cost-utility and cost-effectiveness, and reports of care costs, other economic evidence e.g. cost of illness or burden of disease studies.

Evidence Stream 3 Experience and attitudes (review questions 1, 4-5)

Qualitative, quantitative and mixed-methods evidence exploring experience and attitudes relating to the provision of respite care or short breaks. Studies using recognised methods of data collection and analysis such as surveys, interviews, focus groups, observational techniques, case studies, process and realist evaluations. These may be independent or components of a mixed-methods design.

Evidence Stream 4 UK Policy and guidelines (review questions 1, 6)

All relevant current UK Government policy, clinical guidelines, and NFPO literature will be included.

5.4.1 Data extraction strategy

Data extraction forms will be developed and piloted for each evidence stream, tailored to the type of evidence and the underlying review question. Data will be extracted independently and checked for accuracy by a second reviewer. Disagreements will be resolved through consensus and a third reviewer will be consulted where necessary. Data from multiple publications of single studies will be extracted and reported as a single reference. Where possible, we will attempt to contact authors for missing data.

For all evidence streams, we will extract study aims, methods and population characteristics. We will extract the following detailed information where possible including:

- Publication characteristics: for example, type (peer reviewed), year, country of data collection, dates of study data collection, language, source of funding
- Methods: for example, review, experimental, observational, cross-sectional, case-series, case-report, qualitative (e.g. interviews, focus groups), mixed-methods (e.g. clinical trials with a nested qualitative study, survey and focus groups), duration of follow-up
- Aims, objectives, hypotheses, target audience (policy)

- Intervention description, including programme theory, logic, and design. We will use the TIDieR checklist to extract data (51)
- Participant characteristics: for example, type of CHCNs, duration of the life-limiting/complex health or complex physical disability, 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.

5.4.2 Quality assessment strategy

The methodological limitations of included evidence will be assessed using appropriate tools for each evidence stream. The quality of included evidence will be assessed by one reviewer, and independently verified by a second reviewer. Disagreements will be resolved through consensus and if necessary a third reviewer will be consulted. The outcome of the quality assessment exercise will be tabulated for each evidence stream.

The following tools will be used to assess each type of evidence:

Table 4. Quality assessment tools

Experimental	Randomised controlled trial	Cochrane Risk of Bias (RoB) tool (53)
	Non-randomised controlled trial	Cochrane RoB tool Effective Practice and Organisation of Care (EPOC) adaptations for different study designs (54)
	Before and after study	Cochrane RoB tool (53) or National Institutes of Health (NIH) tool (55)
Observational	Cohort	Critical Appraisal Skills Programme (CASP) for cohort studies (56)
	Case-control	CASP for case control studies (57)
	Cross-sectional	Center for Evidence-Based Management (CEBMa) tool (58)
	Interrupted time-series	Cochrane RoB (EPOC adaptation) (54)
	Case report/case series	Center for Evidence-Based Management (CEBMa) tool (59)
	Economic evidence	CHEERS checklist (60)
Qualitative	Qualitative	CASP for qualitative studies (61)
Mixed-methods	Mixed-methods	Mixed Methods Appraisal Tool (MMAT) (62)
Policy	Policy/guideline document	Appraisal of Guidelines, Research and Evaluation (AGREE II) (63)
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 (64).

5.4.3 Methods of data synthesis

A detailed model illustrating the review design and the types of synthesis to be used within each evidence stream, is shown in Appendix 3. The analysis and synthesis of the different types of evidence is described below. All findings will be discussed by the team in the first instance. It is likely that there will be considerable variability between studies within each evidence stream due to the nature of respite care, specific research methods and the way that outcomes are reported.

5.4.4 *Evidence Stream 1 (intervention effectiveness)*

All quantitative data will be tabulated and grouped by intervention, comparator and outcome where appropriate. Where data allows, meta-analysis will be used to estimate the effects of the intervention for each outcome. Evidence will be included in meta-analyses where the delivery of respite care, data collection methods and populations are similar.

To estimate the heterogeneity among included studies we will use the Chi^2 test ($P \leq 0.10$) to estimate significance and the I^2 statistic to measure magnitude in each meta-analysis, where relevant. We will use the following interpretation thresholds, based on recommendations in the Cochrane Handbook for Systematic Reviews of Interventions (53):

a) 0% to 40%: might not be important
b) 30% to 60%: may represent moderate heterogeneity
c) 50% to 90%: may represent substantial heterogeneity
d) 75% to 100%: considerable heterogeneity.

If we identify substantial heterogeneity (> 50%) (53) we will report this in the text and explore possible causes by using the subgroups specified in Section 5.7.5. It is likely that included evidence will vary by population, specific study design and outcome reporting, but we are also likely to find only a small number of low powered studies. To resolve uncertainty over choice of analysis method we will compare pooled data estimates from both a random-effects model and a fixed-effect model, reporting the mean effect estimate and the confidence interval (CI) around the estimate for both models. We will synthesise and report dichotomous and continuous data separately for a given outcome should the need arise. We will also report end-of-study point estimates and change from baseline scores separately. Where there is sufficient data we will conduct sensitivity analyses based on missing data and risk of bias criteria (randomisation). We will perform the analyses using Review Manager (RevMan) Version 5 (65). However, the team anticipate that meta-analyses may not be possible due to heterogeneity.

5.4.5 *Evidence Stream 2 (health economics)*

All data relating to cost-effectiveness will be tabulated and synthesised narratively, taking into account the following: population size, model of respite care/short break, the comparator, perspective (patient/NHS and social services/society for example), types of costs considered (direct medical and non-medical costs and productivity losses), price year and currency, and outcome measure (QALY, admission avoided, carer's burden, for example), time horizon, discount rate, HRQoL (health related quality of life) instrument used (if applicable).

5.4.6 Evidence Stream 3 (experience and attitudes)

Framework synthesis will be used to synthesise the qualitative evidence. Quantitative data will be synthesised using the methods described in stream 1. An adapted form of framework synthesis will then be used to integrate the narrative findings from the framework synthesis with numerical data from quantitative data (66).

*5.4.7 Evidence Stream 4 (*UK policy and guidelines)

We will conduct content analysis of the documents using a documentary analysis informed approach (67) to construct comparative tables of evidence based on an a priori framework as outlined above. The document analysis will use the eight steps process outlined for textual analysis (68). This approach is an efficient and effective way of gathering extracting and synthesising data from documents.

5.4.8 Sub-group analysis

Where data are reported for subgroups of interest, and if considered appropriate, we will explore differences in outcomes between young adults who have transitioned from paediatric services to adult services, and young adults who developed CHCNs in adulthood (e.g. later development of a condition or following trauma).

The reported prevalence of life-threatening conditions is significantly higher in Black (70.8 per 10,000) and South Asian (31.5 per 10,000) populations, compared to White (25.7 per 10,000) or Chinese/Other (24.4 per 10,000) populations (69). We will consider the differential impacts in relation to gender, socioeconomic status, or ethnicity for example, and where feasible we will conduct sub-group analyses to examine the effects of interventions according to factors in 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 (70).

5.4.9 Overall synthesis

We will use the Framework method for overall synthesis, advocated by the Evidence for Policy and Practice Information and Co-ordinating (EPPI) Centre (71,72). The team will conduct within service type and evidence stream integration of qualitative and quantitative data (72) by juxtaposing evidence in an a priori framework, based on the review questions and policy, to develop themes and sub-themes that will elicit the programme theory of the interventions (models of service), leading to development of the logic models. Team members with expertise in quantitative and qualitative analysis have been assigned to each stream to ensure that there are appropriate skills for synthesis of mixed-methods evidence. Arbitrators are also assigned to each evidence stream to mediate disagreements and uncertainties.

5.4.10 Overall assessment of the evidence

For synthesised findings of effectiveness (Stream 1) two reviewers will independently assess strength of the evidence for all outcomes using the Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach (73). GRADE considers within-study risk of bias, directness of evidence, heterogeneity, precision of effect estimates, and risks of publication bias (74). We will present a summary of findings table using GRADEpro software (73). Evidence is rated as follows:

1. High quality: further research is very unlikely to change confidence in the estimate of effect
2. Moderate quality: further research is likely to have an important impact on confidence in the estimate of effect and may change the estimate
3. Low quality: further research is very likely to have an important impact on confidence in the estimate of effect and is likely to change the estimate
4. Very low quality: we are very uncertain about the estimate.

For synthesised qualitative findings we will use the GRADE-CERQual (Confidence in the Evidence from Reviews of Qualitative Research) approach to evaluate qualitative evidence (75). The CERQual approach makes judgements using four components: methodological limitations of included studies, relevance of contributing studies to the review question, coherence of study findings and adequacy of the data supporting the study findings. Two reviewers will independently assess each CERQual component individually and across the four components. The CERQual levels of confidence are as follows:

1. High confidence: It is highly likely that the review finding is a reasonable representation of the phenomenon of interest
2. Moderate confidence: It is likely that the review finding is a reasonable representation of the phenomenon of interest
3. Low confidence: It is possible that the review finding is a reasonable representation of the phenomenon of interest
4. Very low confidence: It is not clear whether the review finding is a reasonable representation of the phenomenon of interest.

We will tabulate the summary of findings in a CERQual Qualitative Evidence Profile (similar to quantitative findings assessed by GRADE) after using the appropriate quality assessment tools listed above. Evidence not evaluable using the GRADE tools will be reported separately.

We will assess the quality of evidence from policy and guidelines in Stream 4, using the Appraisal of Guidelines, Research and Evaluation (AGREE II) tool (63).

We will develop the overall report using RevMan, the software used by Cochrane (65).

5.5 Conceptual framework

Development will be an ongoing process that builds on Stage 1 and extends across the timeline of the Stage 2 evidence review and into the overarching synthesis. We will develop programme theories and logic models for the different types of respite care using Cochrane guidance (76) and examples of good practice (77,78). Service types will be determined broadly by type, eligibility criteria and target population based on evidence extracted from source papers 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, such as a two week holiday up to the value of X with 4 other patients in a hotel in England). The conceptual framework will be shared and developed with the SG and the PAG to ensure it is appropriate, relevant, and fit for purpose.

6 EXPERTISE AND RESEARCH ROLES

6.1 The review team

Research roles and individual contributions of the review team are detailed below.

Table 5. Research roles

	All team members have contributed to development of the protocol, and will contribute to refining the searches, development of logic models and conceptual framework, overall synthesis, and the preparation of the final report. Each team member has a nominated priority stream which relates to their experience but will work across other streams where needed.
SS	Overall project lead (CI) Lead for development of protocol for publication Arbitrator - study inclusion, quality assessment (Stream 1) Data extraction (Stream 1 and 2) Arbitrator (Stream 3 and 4) Data synthesis (Stream 1 and 2)
KK	Project management Co-chair of the SG with a young adult co-chair Co-ordinating meetings and correspondence All aspects of the knowledge map, Stage 1 and 2 screening and selection, and quality assessment (all streams) Data extraction and data synthesis (Stream 4) Lead for development of conceptual framework
GP	Co-ordinating meetings and correspondence Running searches Retrieval and management of search results All aspects of the knowledge map and evidence review (Stage 1 and 2 screening and selection, data extraction, quality assessment, data synthesis) across all streams
MM	Refining and running searches Retrieval and management of search results Full-text screening and study inclusion Arbitrator– data extraction (all streams)
JD	Stage 1 and 2 screening and selection for the knowledge map and evidence review Quality assessment Data extraction (Stream 3 and 4) Data synthesis (Stream 3 and 4)
JN	Arbitrator– study inclusion, quality assessment (Stream 4) Arbitrator– data extraction (all streams) Data synthesis (Stream 4) Strategic support to the development of conceptual framework
BR	Full-text screening, study inclusion, quality assessment Arbitrator - study inclusion, quality assessment (Stream 3) Arbitrator– data extraction (all streams) Data synthesis (Stream 3)
LB	Full-text screening, study inclusion, quality assessment Data extraction (Stream 3) Data synthesis (Stream 3)
BJ	Full-text screening, study inclusion, quality assessment Data extraction (Stream 2) Data synthesis (Stream 2)
MoB	Full-text screening, study inclusion, quality assessment Data extraction (Stream 3) Data synthesis (Stream 3)
CM	Arbitrator– study inclusion, quality assessment (Stream 2) Arbitrator– data extraction (Stream 2) Data synthesis (Stream 2)

6.2 Steering Group

Invitations were extended to individuals with an in-depth knowledge of care for young adults with CHCNs or the provision of respite care/short breaks, for example those with professional roles in commissioning or delivering services, clinical experts, and representatives from the PAG. The primary purpose of the SG is to advise the review team on all aspects of the systematic review, including the scope of the research, interpretation of results, and dissemination of the research findings. The SG will act as external supervisors, ensuring that the systematic review is conducted in a robust and rigorous manner. The SG will meet on three occasions, and will liaise via email and telephone where necessary.

The SG will be invited to:

- Discuss and comment on the protocol (clarify concepts and definitions, particularly in relation to inclusion criteria)
- Make the review team aware of sources of evidence that may be missing from the published literature
- Inform the review team of current issues in the field that may have relevance to the project, for example current service provision, changes to local or national policies or best practice
- Consider the implication of findings, particularly in terms of service delivery or policy relevance
- Review and comment on drafts of the final report
- Disseminate the findings to relevant audiences

6.3 PPI Advisory Group

Invitations were extended to existing PPI colleagues, and appropriate organisations and individuals to join the PAG, including representation from young people and adults with LLCs and complex physical disabilities, carers, and parents/guardians.

The PAG will meet on 3 occasions throughout the project, with the additional option of working together via telephone, Skype, or email where needed to suit individual preferences and to support the needs of members for whom travel would be challenging due to their healthcare needs. The aim of the PAG is to ensure that the experiences of people accessing respite and short break services, or challenges with accessing services, have input into the review process.

The PAG will be invited to have input into the following:

- Finalising the protocol (e.g. clarifying concepts and definitions, co-writing the Plain English Summary)
- Make the review team aware of sources of evidence that may be missing from the published literature
- Inform the review team of current issues in the field that may have relevance to the project, for example current service provision, changes to local or national policies or best practice
- Interpretation of findings
- Development of logic models and conceptual framework
- Review and comment on drafts of the final report
- Support development of other outputs to share the findings e.g. short videos, animations
- Effectively disseminate the findings to relevant audiences

Structured and focussed activities at PAG meetings will ensure consultation has clear outcomes in line with the needs of the systematic review. All PAG members will receive ongoing support and guidance throughout the project, and will be invited to attend SG meetings to enable representation and appropriate feedback between the two groups.

7 DISSEMINATION AND IMPACT

The team have a dissemination and impact plan which has been developed in partnership with stakeholders and the PAG. We will share the protocol, knowledge map, results of the review, and the conceptual framework with all relevant audiences and stakeholders. Reference libraries of included and excluded evidence from the review will be publicly available from the review website. To maximise pathways to impact, we will disseminate the findings across a range of networks and media, as well as traditional academic mechanisms such as journal articles and conferences. We will work with the PAG to develop other methods of dissemination, for example, short videos or animations to be posted on YouTube and the review website. We will tailor the plan and mechanisms for dissemination for specific audiences according to the key findings of the review and in consultation with the SG and PAG.

8 REFERENCES

1. Together for Short Lives/Marie Curie Cancer Care. Don't let me down Ensuring a good transition for young people with palliative care needs. 2012.
2. Fraser LF, Miller M, Aldridge J, McKinney PA, Parslow RC. Prevalence of life-limiting and life-threatening conditions in young adults in England 2000-2010. Available from: http://www.togetherforshortlives.org.uk/assets/0000/6736/TFSLAdultReport2013Final.pdf
3. Noyes J, Pritchard A, Rees S, Hastings R, Jones K, Mason H, et al. Bridging the Gap: Transition from Children's to Adult Palliative Care [Internet]. 2014. Available from: https://www.bangor.ac.uk/so/bridging-the-gap/Final Report Bridging the Gap between children%27s and adult palliative care 10-14.pdf
4. Worldwide Palliative Care Alliance. Global Atlas of Palliative Care at the End of Life [Internet]. 2014. Available from: http://www.who.int/nmh/Global_Atlas_of_Palliative_Care.pdf
5. Mooney A, Owen C, Statham J, Craig I, Evans B, Lenehan C, et al. Disabled Children: Numbers, Characteristics and Local Service Provision. [cited 2018 Feb 28]; Available from: http://discovery.ucl.ac.uk/1482347/1/Numbers_and_characteristics_of_disabled_children.pdf
6. Equality Act 2010. [cited 2018 Feb 28]; Available from: http://www.legislation.gov.uk/ukpga/2010/15/pdfs/ukpga_20100015_en.pdf
7. NHS. NHS standard contract for specialised rehabilitation for patients with highly complex needs (all ages). [cited 2018 Feb 28]; Available from: https://www.england.nhs.uk/commissioning/wp-content/uploads/sites/12/2014/04/d02-rehab-pat-high-needs-0414.pdf
8. Marchant R, Lefevre M, Jones M, Luckock B. "Necessary stuff": the socal care needs of chldren wth complex health care needs and ther families.
9. Limbrick P. TAC for the 21st Century: Nine essays on Team Around the Child. Clifford: Interconnections; 2009.
10. Rankin J, Regan S. Meeting Complex Needs : The Future of Social Care Meeting Complex Needs. Housing, care Support. 2004;7(3):4–8.
11. Devitt K, Knighton L, Lowe K. Key Data on 16-25 year-olds Transitions, Disadvantage and Crime Young Adults Today.
12. Care Quality Commission. From the pond into the sea. Children's transition to adult health services. Gallowgate; 2014.
13. National Institute for Health and Care Excellence. Transition from children's to adults' services for young people using health or social care services | Guidance and guidelines | NICE [Internet]. NICE; Available from: https://www.nice.org.uk/guidance/ng43/chapter/Recommendations
14. Fraser L K, Lidstone V, Miller M, Aldridge J, Norman P, McKinney P A, et al. Patterns of diagnoses among children and young adults with life-limiting conditions: A secondary analysis of a national dataset. Palliat Med. 2014;28(6):513–520.
15. Remedios C, Willenberg L, Zordan R, Murphy A, Hessel G, Philip J. A pre-test and post-test study of the physical and psychological effects of out-of-home respite care on caregivers of children with life-threatening conditions. Palliat Med [Internet]. 2015 Mar 29 [cited 2018 Feb 28];29(3):223–30. Available from: http://www.ncbi.nlm.nih.gov/pubmed/25634634
16. Together for Short Lives. A Guide to the Development of Children's Palliative Care Services [Internet]. [cited 2018 Feb 28]. Available from: http://www.togetherforshortlives.org.uk/assets/0001/1649/ACT_Guide_to_Developing_Services.pdf

17. Champagne M, Mongeau S. Effects of respite care services in a children's hospice: the parents' point of view. J Palliat Care [Internet]. 2012 [cited 2018 Feb 28];28(4):245–51. Available from: http://www.ncbi.nlm.nih.gov/pubmed/23413759
18. Together for Short Lives. Children's palliative care definitions. [cited 2018 Feb 28]; Available from: http://www.togetherforshortlives.org.uk/assets/0000/4089/CPC_definitions.pdf
19. NHS Choices. Carers' breaks and respite care - NHS.UK [Internet]. 2015 [cited 2018 Feb 28]. Available from: https://www.nhs.uk/conditions/social-care-and-support/breaks-for-carers-respite-care/
20. World Health Organization. WHO Definition of Palliative Care [Internet]. WHO. World Health Organization; 2012 [cited 2018 Feb 28]. Available from: http://www.who.int/cancer/palliative/definition/en/
21. Kirk S, Fraser C. Hospice support and the transition to adult services and adulthood for young people with life-limiting conditions and their families: A qualitative study. Palliat Med [Internet]. 2014 Apr 18;28(4):342–52. Available from: http://journals.sagepub.com/doi/10.1177/0269216313507626
22. Carter B, Edwards M, Hunt A. "Being a presence." J Child Heal Care [Internet]. 2015 Sep 23 [cited 2018 Mar 17];19(3):304–19. Available from: http://www.ncbi.nlm.nih.gov/pubmed/24459101
23. Doug M, Adi Y, Williams J, Paul M, Kelly D, Petchey R, et al. Transition to adult services for children and young people with palliative care needs: a systematic review. Arch Dis Child [Internet]. 2011 Jan 1 [cited 2018 Mar 17];96(1):78–84. Available from: http://www.ncbi.nlm.nih.gov/pubmed/19948663
24. Fraser LK, Aldridge J, Manning S, O'Leary S, Miller M, McCulloch R. Hospice provision and usage amongst young people with neuromuscular disease in the United Kingdom. Eur J Paediatr Neurol. 2011;15(4):326–30.
25. Department of Health. Better Care: Better Lives. 2008 [cited 2018 Mar 19]; Available from: http://lx.iriss.org.uk/sites/default/files/resources/Better care.pdf
26. Mitchell T, Knighting K, O'Brien M R JBR. Short break and emergency respite care: An exploration of options for children and young people with life-limiting conditions when hospice is unavailable. Int J Palliat Nurs. 2016;22(2):57–65.
27. Gatehouse M, Ward H, Holmes L. Developing Definitions of Local Authority Services and Guidance for Future Development of the Children in Need Census. London; 2008.
28. Kelly D, Lidstone V, Edwards J, Griffiths J. Palliative Care for Adolescents and Young Adults. In: Handbook of Palliative Care [Internet]. Hoboken, New Jersey: John Wiley & Sons, Inc.; 2012 [cited 2018 Mar 19]. p. 247–56. Available from: http://doi.wiley.com/10.1002/9781118426869.ch16
29. Knighting K, Mitchell T, O'Brien M, Jack BA. Emergency Care Impact Assessment (ECIA) Project. Final Report. 2014.
30. Together For Short Lives. Transition: A guide for clinical commissioning groups. 2015.
31. Whiting M. Support requirements of parents caring for a child with disability and complex health needs. Nurs Child Young People. 2014;26(4):24–7.
32. Ling J. Respite support for children with a life-limiting condition and their parents: a literature review. Int J Palliat Nurs. 2012;18(3):129–34.
33. Robertson J, Hatton C, Wells E, Collins M, Langer S, Welch V, et al. The Impacts of short break provision on families with a disabled child: an international literature review. Health Soc Care Community [Internet]. 2011 Jul 1 [cited 2018 Mar 17];19(4):337–71. Available from: http://doi.wiley.com/10.1111/j.1365-2524.2010.00977.x
34. National Collaboration for Integrated Care and Support. National Collaboration for Integrated Care and Support [Internet]. [cited 2018 Mar 17]. Available from: https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/198748

/DEFINITIVE_FINAL_VERSION_Integrated_Care_and_Support_-_Our_Shared_Commitment_2013-05-13.pdf

35. Marsh S, Cameron M, Duggan M, Rodrigues J. Young people with life-limiting conditions: transition to adulthood. “Small numbers, huge needs, cruel and arbitrary division of services”. Executive Summary of Phase 1 report for Marie Curie Cancer Care [Internet]. 2011 [cited 2018 Mar 17]. Available from: https://www.mariecurie.org.uk/globalassets/media/documents/commissioning-our-services/past-initiatives/ypt/phase1_young-people-transition-exec-summary.pdf
36. Bamford C, Arksey H, Poole M, Kirkley C, Hughes J, Corner L, et al. Person- and carer-centred respite care for people with dementia : developing methods of evaluating the effectiveness of different models. 2009;1–202.
37. Department for Education and Skills (DfES) and HM Treasury (HMT). Aiming high for children: supporting families. 2007.
38. Pinney A. Understanding the needs of disabled children with complex needs or life-limiting conditions: what can we learn from national data? Exploratory analysis commissioned by the Council for Disabled Children and the True Colours Trust.
39. Foundation for people with learning disabilities. Learning Disabilty Statistics: Effects of being a Carer [Internet]. 2014. Available from: www.learningdisabilities.org.uk/help-information/LearningDisability-Statistics-/187702
40. Malcolm C, Forbat L, Knighting K, Kearney N. Exploring the experiences and perspectives of families using a children’s hospice and professionals providing hospice care to identify future research priorities for children’s hospice care. Palliat Med [Internet]. 2008 Dec 6 [cited 2018 Mar 17];22(8):921–8. Available from: http://journals.sagepub.com/doi/10.1177/0269216308098214
41. Knighting K, Bray L, O’Brien M R, Downing J, Mitchell T, Kirkcaldy A JBA. Short break/respite care services for young adults with life-limiting conditions: A survey of UK children and adult hospices on current provision, collaborative working and future challenges.
42. Children and Families Act 2014 [Internet]. Available from: http://www.legislation.gov.uk/ukpga/2014/6/pdfs/ukpga_20140006_en.pdf
43. Care Act 2014 [Internet]. Available from: http://www.legislation.gov.uk/ukpga/2014/23/pdfs/ukpga_20140023_en.pdf
44. Harden A, Thomas J, Cargo M, Harris J, Pantoja T, Flemming K, et al. Cochrane Qualitative and Implementation Methods Group guidance paper 5: methods for integrating qualitative and implementation evidence within intervention effectiveness reviews. J Clin Epidemiol [Internet]. 2018;97:70–8. Available from: https://doi.org/10.1016/j.jclinepi.2017.11.029
45. Thomas J, Sutcliffe K, Harden A, Oakley A, Oliver S, Rees R, et al. Children and Healthy Eating: A systematic review of barriers and facilitators. EPPI-Centre, Social Science Research Unit, Institute of Education, University of London; 2003.
46. Whitaker R, Hendry M, Aslam R, Booth A, Carter B, Charles JM, et al. Intervention now to eliminate repeat unintended pregnancy in teenagers (INTERUPT): A systematic review of intervention effectiveness and cost-effectiveness, and qualitative and realist synthesis of implementation factors and user engagement. Health Technol Assess (Rockv). 2016;20(16):1–214.
47. Bray N, Noyes J, Edwards RT, Harris N. Wheelchair interventions, services and provision for disabled children: A mixed-method systematic review and conceptual framework. BMC Health Serv Res. 2014;14(1):1–18.
48. Booth A. Clear and present questions: formulating questions for evidence based practice. Cleyle S, editor. Libr Hi Tech [Internet]. 2006 Jul [cited 2018 Mar 19];24(3):355–68. Available from: https://www.emeraldinsight.com/doi/10.1108/07378830610692127
49. Booth A, Harris J, Croot E, Springett J, Campbell F, Wilkins E. Towards a

methodology for cluster searching to provide conceptual and contextual “richness” for systematic reviews of complex interventions: case study (CLUSTER). BMC Med Res Methodol [Internet]. 2013 Dec 28 [cited 2018 Mar 19];13(1):118. Available from: http://bmcmedresmethodol.biomedcentral.com/articles/10.1186/1471-2288-13-118

50. Veritas Health Innovation. Covidence systematic review software [Internet]. Melbourne, Australia; Available from: www.covidence.org
51. Hoffmann TC, Glasziou PP, Boutron I, Milne R, Perera R, Moher D, et al. Better reporting of interventions: template for intervention description and replication (TIDieR) checklist and guide. BMJ [Internet]. 2014 Mar 7 [cited 2018 Mar 19];348:g1687. Available from: http://www.ncbi.nlm.nih.gov/pubmed/24609605
52. Moher D, Liberati A, Tetzlaff J, Altman DG, Group TP. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med [Internet]. 2009 Jul 21 [cited 2018 Mar 19];6(7):e1000097. Available from: http://dx.plos.org/10.1371/journal.pmed.1000097
53. Higgins JPT, Green S. Cochrane Handbook for Systematic Reviews of Interventions. Version 5.1.0. [updated March 2011] [Internet]. Sussex: The Cochrane Collaboration; 2011. Available from: http://handbook.cochrane.org.
54. Cochrane Effective Practice and Organisation of Care (EPOC). Suggested risk of bias criteria for EPOC reviews [Internet]. 2016. p. 1–3. Available from: http://epoc.cochrane.org/sites/epoc.cochrane.org/files/public/uploads/Resources-for-authors2017/suggested_risk_of_bias_criteria_for_epoc_reviews.pdf
55. National Heart, Lung, and Blood Institute (NHLBI). Study Quality Assessment Tools [Internet]. [cited 2018 Jul 13]. Available from: https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools
56. Critical Appraisal Skills Programme (CASP). 12 questions to help you make sense of a Cohort Study How [Internet]. Available from: https://casp-uk.net/wp-content/uploads/2018/01/CASP-Cohort-Study-Checklist.pdf
57. Critical Appraisal Skills Programme (CASP). 11 questions to help you make sense of a Case Control Study [Internet]. Case-Control and Cohort Design Framework. 2018. Available from: https://casp-uk.net/wp-content/uploads/2018/01/CASP-Case-Control-Study-Checklist.pdf
58. Center for Evidence-Based Management (CEBMa). Critical Appraisal of a Cross-Sectional Study (Survey) [Internet]. Web Page. 2016. p. 2014. Available from: https://www.cebma.org/wp-content/uploads/Critical-Appraisal-Questions-for-a-Cross-Sectional-Study-july-2014.pdf%0Ahttps://www.cebma.org/
59. Center for Evidence-Based Management (CEBMa). Critical Appraisal of a Case Study. 2016. p. 10.
60. Husereau D, Drummond M, Petrou S, Carswell C, Moher D, Greenberg D, et al. Consolidated Health Economic Evaluation Reporting Standards (CHEERS) statement. BMJ [Internet]. 2013 Mar 25 [cited 2018 Mar 19];346:f1049. Available from: http://www.ncbi.nlm.nih.gov/pubmed/23529982
61. CASP. 10 questions to help you make sense of qualitative research. 2014.
62. Pluye P, Hong QN. Appraising qualitative, quantitative and mixed methods studies included in mixed studies reviews: The MMAT. Annu Rev Public Health [Internet]. 2014 Mar 18 [cited 2018 Mar 19];35(1):29–45. Available from: http://www.annualreviews.org/doi/10.1146/annurev-publhealth-032013-182440
63. Brouwers MC, Kho ME, Browman GP, Burgers JS, Cluzeau F, Feder G, et al. AGREE II: advancing guideline development, reporting and evaluation in health care. Can Med Assoc J [Internet]. 2010 Dec 14 [cited 2018 Mar 19];182(18):E839–42. Available from: http://www.ncbi.nlm.nih.gov/pubmed/20603348
64. Tyndall J. AACODS Checklist [Internet]. Flinders University. 2010. Available from: /dspace.flinders.edu.au/dspace/
65. Review Manager (RevMan). Copenhagen: The Nordic Cochrane Centre, The

Cochrane Collaboration; 2014.
66. Pluye P, Hong QN. Combining the Power of Stories and the Power of Numbers: Mixed Methods Research and Mixed Studies Reviews. Annu Rev Public Health [Internet]. 2014 Mar 18 [cited 2018 Mar 19];35(1):29–45. Available from: http://www.ncbi.nlm.nih.gov/pubmed/24188053
67. Bowen GA. Document Analysis as a Qualitative Research Method. Qual Res J [Internet]. 2009 Aug 3 [cited 2018 Mar 19];9(2):27–40. Available from: http://www.emeraldinsight.com/doi/10.3316/QRJ0902027
68. O'Leary Z. The essential guide to doing your research project. 2nd ed. Thousand Oaks, CA: SAGE Publications, Inc.; 2014.
69. Fraser LK, Miller M, Aldridge J, Mckinney PA, Hain R. Life-limiting and life-threatening conditions in children and young people in the United Kingdom; national and regional prevalence in relation to socioeconomic status and ethnicity Final Report for Together for Short Lives. 2011.
70. Welch V, Petticrew M, Tugwell P, Moher D, O'Neill J, Waters E, et al. PRISMA-Equity 2012 Extension: Reporting Guidelines for Systematic Reviews with a Focus on Health Equity. PLoS Med [Internet]. 2012 Oct 30 [cited 2018 Mar 19];9(10):e1001333. Available from: http://dx.plos.org/10.1371/journal.pmed.1001333
71. EPPI-Centre. The Evidence for Policy and Practice Information, Social Science Research Unit [Internet]. [cited 2018 Mar 19]. Available from: http://eppi.ioe.ac.uk/cms/
72. Oliver S, Harden A, Rees R, Shepherd J, Brunton G, Garcia J, et al. An Emerging Framework for Including Different Types of Evidence in Systematic Reviews for Public Policy. Evaluation [Internet]. 2005 Oct 24 [cited 2018 Mar 19];11(4):428–46. Available from: http://journals.sagepub.com/doi/10.1177/1356389005059383
73. Cochrane Methods. GRADEing. GRADEPro GDT. 2015.
74. Grade Working Group. Grading quality of evidence and strength of recommendations. Br Med J. 2004;328:1490.
75. Lewin S, Glenton C, Munthe-Kaas H, Carlsen B, Colvin CJ, Gülmezoglu M, et al. Using Qualitative Evidence in Decision Making for Health and Social Interventions: An Approach to Assess Confidence in Findings from Qualitative Evidence Syntheses (GRADE-CERQual). PLOS Med [Internet]. 2015 Oct 27 [cited 2018 Mar 19];12(10):e1001895. Available from: http://dx.plos.org/10.1371/journal.pmed.1001895
76. Noyes J, Hendry M, Booth A, Chandler J, Lewin S, Glenton C, et al. Current use was established and Cochrane guidance on selection of social theories for systematic reviews of complex interventions was developed. J Clin Epidemiol [Internet]. 2016;75:78–92. Available from: http://dx.doi.org/10.1016/j.jclinepi.2015.12.009
77. Anderson LM, Petticrew M, Rehfuess E, Armstrong R, Ueffing E, Baker P, et al. Using logic models to capture complexity in systematic reviews. Res Synth Methods [Internet]. 2011;2(1):33–42. Available from: http://doi.wiley.com/10.1002/jrsm.32
78. Kneale D, Thomas J, Harris K. Developing and optimising the use of logic models in systematic reviews: Exploring practice and good practice in the use of programme theory in reviews. PLoS One. 2015;10(11):1–26.

9 APPENDICES

9.1 Appendix 1: Draft search strategy

The following search terms will be used to search the databases. The initial search will be developed in Medline using MeSH and keywords, then translated into other databases utilising thesaurus terms unique to each resource.

Young adults	Family/carers	Life-limiting	Disability	Respite
MeSH terms				
Young adult Adolescent Adult	Family Caregivers Parents	Palliative Care Palliative Medicine Terminally Ill Heart Failure Neoplasms	Disabled Persons Disabled Children Muscular Dystrophy Muscular Dystrophies Cerebral Palsy Spinal Dysraphism Cystic Fibrosis	Respite Care Hospice Care Hospices Hospice and Palliative Care Nursing Day Care, Medical Night Care Intermediate Care Facilities Terminal Care Holidays
Keywords and phrases				
young adult* young person young people youth* emerg* adult* early adult* child* adj3 transition adj3 adult* adolescen* adj3 transition adj3 adult* teenage* adj3 transition adj3 adult* paediatric* adj3 transition adj3 adult* pediatric* adj3 transition adj3 adult* college student* university student* undergraduate* postgraduate*	famil* carer* caregiver* parent* grandparent* sibling relative* relation*	**advanc*** adj3 disease*/illness*/condition*/disorder*/abnormalit*/impairment*/handicap* **degenerative** adj3 disease*/illness*/condition*/disorder*/abnormalit*/impairment*/handicap* **progressive** adj3 disease*/illness*/condition*/disorder*/abnormalit*/impairment*/handicap* **terminal*** adj3 disease*/illness*/condition*/disorder*/abnormalit*/impairment*/handicap* **genetic** adj3 disease*/illness*/condition*/disorder*/abnormalit*/impairment*/handicap* **congenital** adj3 disease*/illness*/condition*/diso	disabilit* disabled handicap* spastic* multiple disabilities impaired motor skill* spinal cord condition* acquired brain injury multiple trauma neurological condition neuromuscular condition multi-organ disease neurodisabilit* cerebral palsy spina bifida cystic fibrosis encephalopathy*	day away day care day centre/center day program* day service holiday* home support hospice intermediate care night care night-time care* partial hospitalization*/hospitalisation relief care* relief support residential care residential home* residential facilit* respite* short break* short stay* sitting service

		rder*/abnormalit*/impairment/*handicap* **chromosomal** adj3 disease*/illness*/condition*/disorder*/abnormalit*/impairment/*handicap* **neurodegenerative** disease*/illness*/condition*/disorder*/abnormalit*/impairment/*handicap* diminished life expectancy limited life expectancy duchenne dying end of life end stage renal failure heart failure incurable life-limit* life adj3 short* live* adj3 short* life adj3 threaten* limited life expectancy LLC LLI muscular dystroph* neoplasm* oncology palliative poor prognosis serious* adj3 ill* terminal* adj3 ill* terminal* adj3 care* complex health need* early death*		support program* support scheme support service* temporary admission* temporary break* temporary care* temporary relief temporary support short-term admission short-term break* short-term care* short-term relief short-term support time off vacation* care services overnight stay home-based support befriend* service* short-break foster* adult placement scheme* shared care replacement care family support

9.2 Appendix 2: Organisations and charities (grey literature search)

Networks and organisations
Association for Palliative Medicine
European Association for Palliative Care (EAPC).
International Children's Palliative Care Network (ICPCN)
National Institute for Health and Care Excellence (NICE)
NHS CCGs in the UK, Wales, Scotland and Northern Ireland
North West Clinical Commissioning Network
North West Coast Strategic Clinical Networks and Senate
North West Palliative Care Clinical Network
Palliative Care Research Society
Peninsula Cerebra Research Unit for Childhood Disability Research (PenCRU)
Parent Voices Count
Royal College of Nursing
Royal College of Paediatrics and Child Health
Social Care Institute for Excellence (SCIE)
TfSL Regional Action Groups and Transition Taskforce
The Collaboration for Leadership in Applied Health Research and Care, North West Coast (CLAHRC NWC)
The Innovation Agency (IA)
UK local authority websites
Charities
Action Duchenne https://www.actionduchenne.org/
bibic https://bibic.org.uk/
Brain Injury Hub https://www.braininjuryhub.co.uk/
British Heart Foundation https://www.bhf.org.uk/
Carers Trust https://carers.org/
Carers UK https://www.carersuk.org/
Cerebra http://www.cerebra.org.uk/
CHAS https://www.chas.org.uk/how-we-help/family-support/children
Cystic Fibrosis Trust https://www.cysticfibrosis.org.uk/
Disability Rights UK https://www.disabilityrightsuk.org/
Down's Syndrome Association https://www.downs-syndrome.org.uk/
Epilepsy Action https://www.epilepsy.org.uk/
Genetic Disorders UK http://www.geneticdisordersuk.org/
Headway http://www.headway.org.uk/
Hospice UK https://www.hospiceuk.org/
Huntington's Disease Association https://www.hda.org.uk/
MDA https://www.mda.org/
MENCAP https://www.mencap.org.uk/
Motor Neurone Disease Association https://www.mndassociation.org/
Motor Neurone Disease Scotland (MND Scotland) https://www.mndscotland.org.uk/
MS Society https://www.mssociety.org.uk/
Muscular Dystrophy UK http://www.musculardystrophyuk.org/

My Life https://www.my-life.org.uk/
Rainbow Trust https://rainbowtrust.org.uk/
Reach http://reach.org.uk/
SCOPE https://www.scope.org.uk/
Shine https://www.shinecharity.org.uk/
Spinal Injuries Association https://www.spinal.co.uk/
SWAN UK http://www.undiagnosed.org.uk/
Together for short lives http://www.togetherforshortlives.org.uk/

9.3 Appendix 3: Review Design and Synthesis Methods Model

Review Questions	Streams	Study type	Selection of evidence for in-depth review data extraction	Synthesis of evidence according to type using appropriate methods	Overarching narrative synthesis and programme logic models for interventions
What is the effectiveness and cost effectiveness of different types of formal and informal respite care and short break provision? What is the economic impact of respite care and short breaks?	Stream 1 Intervention Effectiveness	RCTs and quasi-RCTs	Searches Screening Study inclusion/exclusion Appraisal of quality or risk of bias Data extraction Confirm typology and different interventions (models of service)	Meta-analysis Narrative summary	Overarching narrative synthesis of qualitative and quantitative evidence including articulation of programme theories Logic model development for interventions (models of service) to show the different designs, inputs, processes and intended outcomes for various stakeholders
		Other experimental or quasi experimental studies			
	Stream 2 Health Economics	Cost-utility, cost-effectiveness		Meta-analysis Narrative summary	
		Other economic studies			
What are service users and providers views of current service provision and the need for new services? What are the facilitators and barriers to providing, implementing and using respite care and short breaks, taking into account the different perspectives of service users, family members and providers?	Stream 3 Experience and Attitudes	Quantitative (e.g. questionnaire surveys)		Narrative summary (quantitative data)	
		Qualitative studies, and qualitative data, e.g. from open ended survey questions		Thematic synthesis (qualitative data)	
		Mixed-method studies (e.g. case studies and process evaluations)		Mixed-method matrix for integration of quantitative and qualitative evidence	
What are the current UK policy and guidance recommendations for the provision of respite care and short breaks?	Stream 4 Policy and Guidelines	Clinical guidelines		Framework synthesis	
		Government policy			
		Other			