The engagement of young people in their own advance care planning process: a systematic narrative synthesis

Ben Hughes, Professor Mary R. O'Brien, Dr Anita Flynn, Dr Katherine Knighting

All members of the research team are from Edge Hill University

Corresponding author:

Ben Hughes
Faculty of Health and Social Care
Edge Hill University
St Helens Road
Ormskirk
Lancashire
L39 4QP

Tel: 01695 654 354
Email: Ben.Hughes@edgehill.ac.uk

Running title: Engagement of young people in advance care plans

Word count: 5,352

Abstract

Background:

An increasing number of young people are living with life-limiting conditions. Current research about advance care planning for young people indicates differing experiences for those involved. Understanding how far young people are engaged in their own advance care plan is important to shape future practice and facilitate young people's wishes.

Aim:

To identify and assess the current evidence to determine the barriers and facilitators to the engagement of young people in their own advance care planning process.

Design:

A systematic narrative synthesis according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. Study quality was assessed using a quality assessment framework[1] previously used in similar research.

Data sources:

CINAHL Complete, MEDLINE, PubMed, and PsycINFO were searched for articles published between 1st January 1990 and 31st October 2017. Grey literature was searched using Google Scholar and Open Grey.

Results:

Most studies related to the engagement of young people were conducted in hospitals or other institutions. Research reported the aim to include young people in their own advance care planning, but also potential barriers to engagement. Barriers include poor communication, conflict within relationships of those in the planning process, and patchy education and training for healthcare professionals. Some existing studies are characterised by a lack of rigorous, high quality, research, limiting their impact.

Conclusion:

Irrespective of setting, engagement of young people would benefit their advance care planning. More detailed, high-quality research is needed to understand the extent of the barriers to young people's engagement in their own advance care plan and how to facilitate their involvement.

Keywords

Advance care planning, systematic narrative synthesis, adolescent, young people, young adult, patient participation

Key statements

What is already known about the topic?

- There are varying experiences for young people engaged in advance care planning
- It is unclear how far young people are engaged in their own advance care planning process

What this paper adds

- This study demonstrates that the aim is to often include young people in their own advance care planning process

- Engaging young people in their own advance care planning process would benefit the standard of care they receive

- Communication, relationships, and the availability and access to training for healthcare professionals, can act as either a barrier or a facilitator to the engagement of young people in their advance care plan.

Implications for practice, theory or policy

- The alignment of policy and practice can both facilitate young people's engagement, and empower them, in their own advance care planning.

- A collaborative approach to advance care planning would help facilitate the engagement of young people in the process.

Introduction

The development of advance care planning

From the late 1990s, the Gold Standards Framework (GSF)[2] has supported a systematic, whole systems, evidence-based approach to improving end-of-life care (EoLC) for all patients.[2,3] Advance care planning (ACP) is one element within the GSF which aims to nurture communication and develop a consistent approach between practitioners[4] by using a formal process to provide high quality EoLC. Advance care plans (ACPs) perform several functions,[5] such as: the opportunity to consider aspects of loss of self at the end-of-life; to acknowledge the prospect of death and negotiate personal preferences around future treatment; to make clear personal preferences

around clinical interventions and personal possessions in the event that capacity for decision-making is lost; and discuss the delegation of future care in the event that capacity is lost.

ACP has been practised for a number of years in the United States, Canada and Australia and is based on the preservation of personal autonomy in decision-making.[6] As an example of patient participation, ACP encourages patients to be involved in their own care.[7] The United Kingdom's first End of Life Care Strategy set out the use of ACP for adults.[8] The resulting Liverpool Care Pathway (LCP) became the standard and most widely-used version of ACP. However, due to a number of high profile cases and media reports between 2009 to 2012,[9] it was finally phased out in 2014.

Advance care planning for young people

There is a misconception that ACP is solely used for terminally ill elderly people.[10,11] Yet an increasing number of young people with complex, life-limiting conditions (LLCs) could benefit from its use.[12] Over 49,000 young people in the UK currently have a LLC or Life-Threatening Condition (LTC).[13] Furthermore, 70% of young people's deaths in England are complicated by problematic LLCs.[12] An understanding of the experiences and level of engagement of this population in their own care planning is important to delivering palliative care.

For this review, young people are those aged 13-24 years. This term corresponds with the definition of a 'young adult' as given by Medical Subject Headings (MeSH) (US National Library of Medicine 2017). This definition also standardises existing age ranges used by children's hospices in the UK.

The lack of a clear and consistent definition of ACP makes it both difficult to identify within the literature and hard to translate theory into practice.[14] Adult ACP focuses on recognising and communicating values, life goals and preferences with specific regard to future medical care.[15] However, advance care planning for young people (ACPfYP) simply records advance wishes of the young person and/or their parent/carer.[16,17] This definition has been developed to include actions that should be taken when the young person's condition becomes unstable, deteriorates or they develop potentially life-threatening complications of their illness.[18]

ACPfYP was developed from 2010 to help young people and their parents/carers communicate their wishes, as well as helping with the grieving process and support around the death of the young person.[19] However, palliative care resources are focused more on provision for adults than provision for young people.[20] Consequently, most research currently focuses on ACP for adults and there is limited research into the engagement of young people in their own ACP.

The implementation of ACPfYP during palliative care is a key contributor to the success of effective communication.[21] This, in turn, can help determine high quality care.[22] Research suggests there are varying experiences for those using ACPfYP due to the complex needs of young people, misunderstanding of healthcare professionals (HCPs) around how and when to implement them[23] and failure to keep parents/carers informed of the process of ACPfYP.[24] Additionally, as with adult ACP, there is no single standard national documentation used for young people.[25] As a result, it is currently unclear how far young people engage in their own ACP. A first step towards improving

the consistent use of ACPfYP, with the involvement of young people, is to understand the barriers and facilitators for young people's engagement in their own ACP. This review is a timely summary of existing research to benchmark current understanding of these key areas.

Method

Review question

What are the factors that may facilitate or mitigate engagement of young people in their own ACP?

To help recognise barriers and facilitators to the process of engaging young people in their own ACP, this synthesis has three key objectives to identify key stages of the process and how this impacts on the engagement of young people:

1. to explore who is involved in the advance care planning process;
2. to explore when the process is started; and
3. to explore how key participants (young person, their parents/carers and healthcare professionals) are engaged in the process and how this facilitates or mitigates engagement of the young person.

Review design

Existing evidence about young people's engagement in their ACP is diverse and includes both qualitative and quantitative studies. Therefore, an approach to help synthesise the literature was required. Narrative synthesis was the most appropriate approach to integrate these different studies because its rigourous nature is suitable

to facilitate the synthesis of heterogeneous studies.[26,27] The definition of terms used in

the review is in Table 1.

Table 1 - Definition of terms

Abbreviation	Term	Definition
ACP	Advance Care Planning	A voluntary discussion-based process between an individual, their care providers and those close to them, about future care where a future loss of the patient's capacity is anticipated.[30]
ACPs	Advance Care Plans	The individual instance of an advance care plan, which is to be distinct from the overarching ACP process.
ACPfYP	Advance care plans for young people	An advance care plan which is implemented for someone aged 13-24 years.
EoL	End-of-life	The period of time before death, usually considered to be the last 12 months of life.
EoLC	End-of-life Care	Care that helps people with advanced, progressive, incurable illness to live as well as possible until they die.[31]
GSF	Gold Standards Framework	Developed from 1998, the GSF supports a whole systems approach to improving end-of-life care (EoLC) for all patients.[3]
HCPs	Healthcare Professionals	Professionals who: study, diagnose, treat and prevent human illness, injury and other physical and mental impairments; advise on or apply preventive and curative measures, and promote health; conduct research and improve or develop concepts, theories and operational methods to advance evidence-based health care; and may also supervise other health workers.[30]
LLCs	Life limiting conditions	Conditions which cause progressive deterioration, for which there is no reasonable hope of cure and from which a child or young person will die.[22]
LCP	Liverpool Care Pathway	An approach to care, including a complex set of interventions, which aimed replicate the hospice standard of case within hospitals.[33]
LTCs	Life-threatening conditions	Conditions in which curative treatment may be feasible but can fail.[33]

-	Palliative Care	An active and total approach to care, from the point of diagnosis or recognition, which continues through to death and beyond.[31]
-	Parallel Planning	Plans are made for life, while also allowing for a deterioration in condition, or even death.[31]
-	Young Person	Someone aged 13-24 years.[34]

Search strategy

Database searches

The databases of Cumulative Index to Nursing and Allied Health Literature (CINAHL Complete), MEDLINE (Medical Literature Analysis and Retrieval System Online), PubMed, (Public Medline) and PsycINFO (Psychological Information Database) were searched for articles published from 1st January 1990 to 31st October 2017. Grey literature was searched using Google Scholar and Open Grey. Citation searching and reference list checking was performed on all identified papers.

Searches included all existing literature and grey literature to present a broad and balanced reflection of the involvement of young people in their ACP. Therefore, the review included both qualitative and quantitative studies and incorporates all study types identified, including systematic reviews, cohort studies, case studies, interviews, randomised control studies and retrospective studies.

Database search terms

Medical subject headings (MeSH), or a thesaurus, were employed to conduct the search, where possible (see Table 2). The Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA)[35,36] guidelines were followed for this review, as in similar reviews[27] .

Table 2 - Search terms used in database searches

1. Advance care plan* **OR** pACP **OR** ACP **OR** wishes document **OR** wishes **OR** My Choices

   AND

2. Paed* **OR** Child* **OR** Young person **OR** Adoles*]

3. End of life **OR** Palliative*

Selection criteria

Studies were selected for review based on clear inclusion and exclusion criteria (see Table 3).

Table 3 – Inclusion and exclusion criteria for study selection

Inclusion criteria	Exclusion criteria
• Sources written in English. • Studies conducted at any location or place of care. • Studies including evidence about the engagement of young people (aged 13-24) in their own advance care planning. • Sources published between 1st January 1990 and 31st October 2017.	• Sources published before 1990. • Sources written in languages other than English. • Studies focused on advance care planning for adults (aged over 24 years) and children (aged under 13 years).

Data extraction and method of synthesis

The search strategy was adopted from similar previous research[27] and adheres to the following narrative synthesis guidelines recommended by Popay et al.[37]

1. The first stage was to conduct a preliminary analysis of the literature to identify relevant sources. This was a two-step approach to identify and then screen the literature:

    i. Searches were conducted using the databases identified above to locate abstracts and, where possible, titles of studies containing the search terms. This generated a list of potential studies to include in the synthesis. Results were then screened to remove duplicates and ensure that relevant studies were included in the synthesis. BH read and assessed the remaining studies for eligibility. This process allowed a further selection and exclusion of studies to take place. Studies which did not meet the inclusion criteria set out above were removed.

    ii. The title and abstract of remaining studies were then screened and studies not meeting the criteria were excluded. The full text of the remaining 25 articles was assessed for eligibility by BH. Over 50% (n=15) of these papers were shared within the research team (MoB, AF, KK) for independent eligibility assessment and to increase the rigour of the study. Disagreements about study inclusion at each stage of study selection were resolved through discussion within the team.
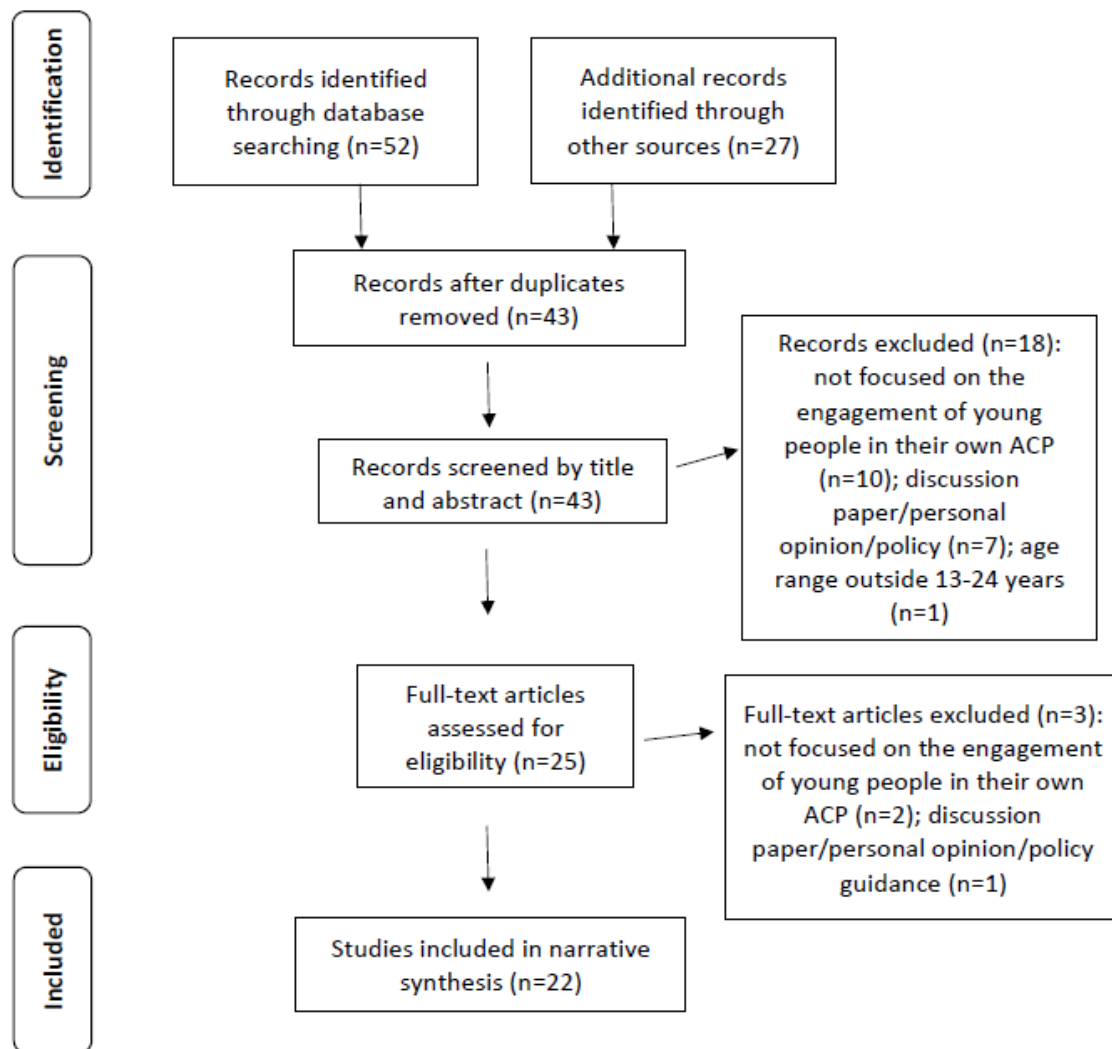
2. The second stage was to explore the relationships between the results from the studies. Data from the studies were extracted and presented in a Characteristics of Included Studies table, before being thematically analysed by BH and discussed within the research team. This process ensured consistency, clarity and transparency within the data extraction and management process. The selected characteristics were based on those identified in previous research into ACP.[38] The table included the following information about each publication: study details; aims and objectives; study design; participant characteristics; country; setting; key findings; theme(s); included/excluded and quality assessment score.

3. Assessment of the robustness of the synthesis was carried out by the research team at each step of the process. A narrative synthesis approach was used to present themes found as it is well-suited to synthesising evidence from studies that are heterogeneous in method.[37]

The review has been registered with PROSPERO (PROSPERO registration number: CRD42017079823).

A total of 22 articles met the inclusion criteria and were included in the synthesis (see Figure 1 – PRISMA flow diagram).

Figure 1 - PRISMA flow diagram

Identification

Records identified through database searching (n=52)

Additional records identified through other sources (n=27)

Screening

Records after duplicates removed (n=43)

Records screened by title and abstract (n=43)

Records excluded (n=18): not focused on the engagement of young people in their own ACP (n=10); discussion paper/personal opinion/policy (n=7); age range outside 13-24 years (n=1)

Eligibility

Full-text articles assessed for eligibility (n=25)

Full-text articles excluded (n=3): not focused on the engagement of young people in their own ACP (n=2); discussion paper/personal opinion/policy guidance (n=1)

Included

Studies included in narrative synthesis (n=22)

Assessment of quality

Hawker et al.'s[1] assessment framework (Appendix 1) and scoring system (Appendix 2) were utilised to assess the quality of the studies included in the synthesis. This approach mirrors that taken in previous similar studies where either palliative care research was undertaken, a systematic narrative synthesis was conducted, or a combination of the two was presented.[27] The assessment framework allows literature to be scored (9 very poor; 36 very good) to indicate the methodological rigour of each study.[1] This scoring process gives a clear indication of the strengths and weaknesses

of each study and so provides clarity, transparency and rigour in the quality assessment process.[1]

Overview of the studies

Publication dates of the studies ranged from 2004 to 2017. Studies were heterogeneous, with the majority being quantitative (n=15)[24,39-51]; three were qualitative method [52-54] and the remaining studies (n=4)[53,55-57] used a mixed methods approach. The 22 studies included were primarily from the United States (US) (n=14),[40,42-44,47-50,52,56-58] with further studies coming from the United Kingdom (n=4),[24,39,54,55] France,[41] Germany,[59] Japan.[51] The remaining study, a systematic review, included research from the US and Australia.[46]

Study settings included: children's services/hospitals/clinics/medical centres (n=11);[39-42,44,45,48,49,52,54,55] cancer centres/institutes (n=2);[50,58] and universities (n=2).[43,57] Other research was conducted within National Health Service (NHS) Trusts;[24] via a national board of haematologists;[51]; and a range of settings: care settings only,[48,59] a combination of care and non-care settings;[46] paediatric hospitals and HIV clinics;[47] and a cancer institute and a university.[53]

Participants varied in their diagnosis: cancer and/or HIV/AIDS (n=9)[40,44,47-49,51,53,58] LLCs or LTCs (n=6);[24,39,41,42,46,54] chronic/severe/serious illness (n=3);[50,56,59] and cystic fibrosis (n=1).[52] No specific condition was diagnosed in the young people in the remaining studies (n=3).[43,55,57]

Similarly, young people's knowledge of their diagnosis varied between studies: it was not mentioned in half of the studies (n=11);[24,39,41,42,44,46,47,53-55,59] young people were aware of their diagnosis in a third of the studies (n=7);[40,48-50,52,58] in a small number of studies (n=2),[51,56] at least some participants were aware of their diagnosis, while others were unaware; and the young people did not have a diagnosed condition in the remaining studies (n=2).[43,57]

All participants were English-speaking and, where race/ethnicity was acknowledged, they identified across a range: Black, African-American, Hispanic/Latino, Asian, Black, White/Caucasian, American Indian/Alaskan, Biracial, Asian/Pacific Islander. Educational attainment and income level was mixed across those studies in which these factors were identified.[40,43,44,47-49,57] The male/female split between young people was fairly equal in most studies.

Themes

Analysis produced four themes: how ACP works in practice; communication; training and education; and relationships; presented in the narrative below. The 22 studies are summarised in Table 3.

Table 3 – Data extraction and study quality

Characteristics of Included Studies Table

Study details	Aims and objectives	Study design	Participant characteristics	Country	Setting	Key findings	Theme(s) of included papers	Included/excluded (Assessment Framework score for included studies)
Keeping all options open: Parents' approaches to advance care planning[54]	To investigate how parents of children and young people with LLCs approach and experience ACP	Open ended, semi-structured interviews	(i) Parents whose child was currently receiving services from the palliative care team (n=28) (ii) Bereaved parents whose child had received care from the palliative care team and had died 6- 10 months previously (n=29) A child population aged 0- 19 years with a diverse range of life-limiting conditions, ethnic and socio-economic backgrounds.	UK	Unidentified specialist palliative care service	Most decisions were made relatively late in the child's illness It is questionable whether the parents in our study would have welcomed earlier discussions Parents need more support and more information about the aims of ACP to help make decisions	ACP in practice • How young people are engaged in ACP discussions Relationships • The dynamics in relationships between young people and their parents • Highlighted the importance of a trusting relationship in the ACP process	Included The focus of the study is about engaging parents as well as young people in ACP Hawker et al, framework score: (30/36)

Was there a plan? End-of-life care for children with life-limiting conditions: a review of multi-service healthcare records[60]	To compare documented EoL planning with published children's palliative care standards, across a range of children's healthcare services and to assess the impact on practice of written guidelines to support EoL care planning	A manual retrospective review of healthcare records	Children with a diagnosed LLC or LTC, who had died before the age of 18 years, between October 2008 and March 2010, within a defined geographical area served by one or more of the participating services (n=114 records relating to 48 children; 24 boys and 24 girls) A range of LLCs	UK	Unspecified services in the West of England	Documented EoL planning varies between services Professionals can help prepare staff to engage families in these conversations	ACP in practice • Varies between services Training/education • Failures in ACP may be related to the need for additional training and access to documentation Relationships • HCPs have an important role to play in ACP discussions	Included The focus of the study is about engaging young people in the ACP process Hawker et al, framework score: (27/30)
Service evaluation of an advance care plan tool[55]	Investigate how accurately the documentation was being completed Ascertain the appropriateness of the tool from the family and staff perspective Discover the extent to which families were able to agree care at the end of life,	Mixed methods	ACP document analysis from 3 research sites Parents (n=4) from 2 research sites Focus groups with unspecified professionals and lead nurses (n=17) No information about the breakdown of gender, ethnicity or	UK	Children's services in East Kent	Inconsistent structure and formatting of ACP documentation, leading to ambiguity in their use HCP knowledge of the young people and the ACP process is important There needs to be a state of awareness and	ACP in practice • Issues around the format of documentation • Timing of ACP needs to be considered • Young people should be involved in ACP discussions Communication • Barriers to families wanting to engage	Included Focus is on engagement of families rather than the engagement of young people Hawker et al, framework score: (32/36)

| | | | | | | readiness for the ACP process to engage parents

ACPs may be inappropriate in some circumstances e.g. conflict with religious beliefs

ACP can be positive if used proactively, is family-driven and is introduced in the home environment

Timing of ACP is important e.g. not when the young person's condition is unstable | Training/education
• Current training not always consistent

Relationships
• Some tension in relationships | |
|---|---|---|---|---|---|---|---|---|
| | including resuscitation decisions

Identify staff perceptions of training received regarding application of the tool

Identify and build on the strengths, and make recommendation for improvement | | | socio-economic groups | | | | | |
| Including the Perspective of the Adolescent in Palliative Care Preferences[52] | To present issues and challenges that HCPs encounter and to describe how communication can be facilitated between the patient, family and HCPs | Case study | The case is an 18-year-old woman diagnosed with cystic fibrosis as an infant. Her disease progression has resulted in more frequent and longer hospitalisations | US | Children's Mercy Hospitals and Clinics in Kansas City, Missouri | Communication tools:
• can facilitate young people's perceptions, beliefs, values, preferences and goals, as well the success of available coping mechanism | ACP in practice
• How ACP works for the young person in the study

Communication
• Can help understand young people and the care they need
• Can help young people develop skills to convey their own care | Included

Discusses the engagement of young people within the family context

Hawker et al, framework score: (21/36) |

							• can help initiate difficult conversations • may initiate thinking about personal and health care goals	treatment decisions	
Acceptability of Family-Centered Advanced Care Planning for Adolescents With HIV[40]	To determine the appropriateness of engaging adolescents with chronic or life-limiting illnesses in advance care planning	Randomised control trial	Young people aged 14-21 years Participants were aware of their HIV diagnosis; a documented IQ score <70; no documented diagnosis of severe depression, suicidal/homicidal ideation or psychosis; and able to understand English	US	6 unspecified US urban hospitals with high AIDS rates	Family-centred ACP enabled worthwhile conversations, while simultaneously eliciting intense emotions Evidence of the feasibility, acceptability, and safety of ACPfYP	ACP in practice • Families want to hear their own child's voice • Resilience of young people in ACP discussions Communication • FaCE enabled high quality EoL conversations between families and their teens	Included Identifies attitudes of young people to ACP Hawker et al, framework score: (29/36)	
Advance care planning and place of death in a paediatric palliative care unit in France[41]	To compare two periods to evaluate the impact of ACPs Identify the challenges and benefits of developing advance care plans with the	Retrospective study	No specific information provided The article was a general discussion of the experiences of the authors at their paediatric palliative care unit	France	Paediatric palliative care unit	Advance care planning is crucial to involve families of paediatric patients in decision-making and help them understand that they can have their say in what happens to their child in the final phase	ACP in practice • Starting ACP discussions early is beneficial Communication • ACP discussions can be difficult but are beneficial Relationships	Included Includes the role of young people within the family context in the ACP process Hawker et al, framework score: (21/36)	

	families of paediatric patients							• Potential for conflict	
Barriers to Conducting Advance Care Discussions for Children With Life-Threatening Conditions[42]	To identify barriers to conducting advance care discussions for children with LTCs	Survey	Physicians (n=107) and nurses (n=159). Participants included all ICU and oncology attending physicians, fellows, hospitalists, nurses, and advance practice nurses.	US	The medical/ surgical ICU, medicine ICU, cardiac ICU (CICU), and oncology ward at Children's Hospital Boston; and the outpatient oncology service at the Dana-Farber Cancer Institute	Clinicians perceive parent prognostic understanding and attitudes as the most common barriers to conducting advance care discussions. Educational interventions aimed at improving clinician knowledge, attitudes, and skills in addressing these barriers may help HCPs overcome perceived barriers.	Communication • New resuscitation order will be developed to communicate overall goals of care Training/education • HCPs' ability to discuss EoLC with families is a barrier to engagement in ACP	Included Although the focus of the study was HCPs, the research identified the impact of their knowledge and skills on the process of ACPfYP Hawker et al, framework score: (29/36)
The Importance of Inter-generational Communication in Advance Care Planning: Generational Relationships among Perceptions and Beliefs[43]	Explores triadic inter-generational perceptions of family members' beliefs and behaviours that often impact an individual's willingness to engage in advance care planning	An online survey, covering openness about death; death anxiety; perceived knowledge about surrogate decision-making; and ACP self-efficacy	Undergraduate students (n=567) comprising men (n=173) and women (n=394) Aged 18-88 The majority of participants in the project were white (n=426), followed by Hispanic (n=39) and Asian (n=28).	US	Surveys were completed online	The need to treat advance care planning as a family communication issue	Communication • Effective communication should be targeted for effective ACP Relationships • Importance of effective family relationships and negotiating conflict	Included This study is about family engagement in the ACP process and includes the perspective of young people Hawker et al, framework score: (29/36)

			African Americans and those who identified as Middle Eastern each comprised 10 participants. The majority of respondents identified as Catholic (n=179) or Protestant (n=234); nondenominational Christian (n=64) and no religion (n=44). The remainder of participants identified as Jewish, Hindu, Mormon, Muslim, and Buddhis				• A family-centred approach might encourage people to engage in ACP	
An Exploratory Survey of End-of-Life Attitudes, Beliefs, and Experiences of Adolescents With HIV/AIDS and Their Families[44]	To examine congruence in HIV positive adolescents and their families' thoughts about death and dying.	Survey administered within a randomised control trial	Medically stable, English-speaking adolescents aged 14-21 years with HIV/AIDS and their families (n=48) Adolescent participants: female (n=15) and male (n=9) Family participants: female (n=19) and male (n=5)	US	Children's National Medical Center and St. Jude Children's Research Hospital, Michigan, Washington	The timing of EoL conversations and adolescents' EOL needs and wishes are not known by their families. Families need help initiating EoL conversations to assure that their adolescents' EOL wishes are known to them	Communication • Needs to be improved with young people	Included This study includes the engagement of young people and their inclusion of their wishes in their EoL decisions. Hawker et al, framework score: (30/36)

			Black/African American (n=41) and only 3 participants (all family members) were educated to university level. Nearly half of the participants (n=21) did not have any qualifications. Nearly half of the family groups (n=10 out of 24) identified themselves as living at or below the federal poverty level					
Advance Care Planning: Challenges and Approaches for Pediatricians[24]	To review advance care planning for children with life-threatening or life-limiting conditions (LTLLCs) in our local area	Retrospective case note review	Young people aged 0-18 years (n=20) who were diagnosed with a LLC or LTC Died between October 2008 and March 2010, which was caused by the progressive disease/underlying diagnosis (n=11) or respiratory problems (n=7)	UK	Two NHS Trusts in Bristol	The numbers of children living with LCCs or LTCs are estimated to be higher than previously thought Advancing technologies can sustain life in ways that were previously impossible The best interests of the child should be at the forefront	ACP in practice • Varying standards of ACP Communication • Using prompts can help initiate conversations about ACP • Communication skills are key	Included This study is about family engagement in the ACP process and includes the engagement of young people Hawker et al, framework score: (24/36)

							of the EoL decision-making process There are clear benefits of ACP	
Adolescent End of Life Preferences and Congruence With Their Parents' Preferences: Results of a Survey of Adolescents With Cancer[58]	To examine baseline congruence between the self-reported needs of adolescents with cancer for EoLC and their families' perception of those needs To better understand how adolescents with cancer approach EoL issues Practitioners' experiences with the study and their thoughts regarding EOL care for themselves and their patients	Survey	English-speaking, age 14-21 years, and were diagnosed with cancer (n=17) and their family member (n=17) Mean age of the adolescents was 16 years (range 14-21; 12 patients (71%) were less than 18); 63% were male, and 58% were African-American	US	Adolescents and families in the intervention arm of the FAmily CEntered Advance Care Planning (ACP) for Teens with Cancer (FACE- TC) study	Adolescents with cancer were comfortable discussing EoL, and the majority preferred to talk about EoL issues before they are facing EoL Important facets of adolescents' EoL wishes were not known by their families, reinforcing the importance of eliciting individual preferences and engaging dyads so parents can understand their children's wishes	ACP in practice • Need improved information about hospice services • Not always congruence the family about location of death Training/education • Improved communication between HCPs and families	Included Identifies that young people can/should be engaged with planning their own care Hawker et al, framework score: (26/36)
Pediatric Advance Care Planning: A Systematic Review[46]	To assess current practices, effects, and perspectives of pACP	Systematic review	5 qualitative and 8 quantitative studies. Only 3 pACP programs were identified, all from the United States	US	A range of settings: Inpatient/ outpatient	There are few systematic pACP programs worldwide and none in Europe	ACP in practice • Reluctance to engage in ACP discussions if the prognosis was poor	Included Review included attitudes of young people in the EoL/ACP process

					and/or clinic (n=5) ICU (n=1) School (n=1) Unspecified (n=4)	Future research should investigate the needs of all stakeholders, particularly HCPs		Hawker et al, framework score: (33/36)
Participants ranged in age from 2 months to 37 years old								
Pediatric advance care planning from the perspective of health care professionals: A qualitative interview study[59]	To investigate attitudes and needs of HCPs with regard to pACP To investigate attitudes, barriers, and benefits as well as requirements for pACP from the view of HCPs	Qualitative interview study	17 HCPs (physicians: n=9; nurses: n=6; social professionals n=2) representing a range of paediatric disciplines An average age of 46 years and an average of 21 years' experience Male (n=6); female (n=11)	Germany	Hospital care (n=7) Outpatient care (n=8) Care facility (n=4) Some participants were working in more than one care setting in Munich and surrounding areas	There is a need for paediatric ACP There are several barriers to its implementation	ACP in practice • Problems related to ACP are about HCP discomfort and uncertainty • Potential barriers and facilitators to ACP are identified • Evidence from adult ACP may help to inform the development of ACPfYP Training/education • Focus for HCPs' training is identified	Included Identified the engagement of young people from HCPs' point of view Hawker et al, framework score: (33/36)
A randomized clinical trial of adolescents with HIV/ AIDS: pediatric advance care planning[47]	To determine if paediatric advance care planning (pACP) increases adolescent/ family congruence in end-of-life (EOL) treatment preferences longitudinally	Randomised clinical trial	Adolescents aged 14-21 years with HIV/AIDS and their families (n=105 dyads). Adolescents- Male (n=29); female (n=25)	US	6 unspecified paediatric hospital-based HIV clinics, located in high mortality urban areas in the Southern US	A structured pACP intervention was effective in: Increasing congruence in EOL treatment preferences between adolescents and their families	ACP in practice • ACP is more effective when intervention is targeted • Targeted ACP positively influenced relationships and communication	Included Review included attitudes of young people in the EoL/ACP process Hawker et al, framework score: (35/36)

			Age range=14-20 years; mean age=18 years; Black African-American (n=50); Hispanic/Latino/non-African (n=4). No qualifications from high school (n=29) At or below the federal poverty line (n=28) One-third had an AIDS diagnosis.			Engaging adolescents/families to begin these conversations early and to continue these conversations over time Demonstrating safety e.g. such topics as using a seat belt and having a smoke detector in the home		
A Longitudinal, Randomized, Controlled Trial of Advance Care Planning for Teens With Cancer: Anxiety, Depression, Quality of Life, Advance Directives, Spirituality[48]	To test the feasibility, acceptability and safety of a paediatric advance care planning intervention	Randomised controlled trial	Adolescent (age 14-21 years)/family dyads (n=30) with a cancer diagnosis Young people: Male (n=18), female (n=12); Race: Asian (n=2); Black (n=13); White (n=15) A range of diagnoses: Leukaemia (n=14); Lymphoma (n=2); solid tumour (n=6); brain tumour (n=8) The majority (n=24) had no educational qualifications	US	Unidentified large urban children's hospital in-patient unit, outpatient clinic and patients' home	Family-Centered Advance Care Planning for Teens With Cancer demonstrated feasibility and acceptability. Courageous adolescents willingly participated in highly structured, in-depth paediatric advance care planning conversations safely	ACP in practice • Structured ACP can improve participation in the ACP process and communication; and reduce stress and anxiety	Included Study discusses ACPfYP with the target age group Hawker et al, framework score: (35/36)

			Family members: Male (n=2), female (n=28) Race: Asian (n=1); Black (n=13); White (n=13); declined (n=1) No high school diploma or were in high school (n=24) An income >300% federal poverty line (n=14),with only 4 families reporting an income equal to or below the federal poverty line					
Family-Centered Advance Care Planning for Teens With Cancer[45]	To examine the efficacy of family-centred ACP	Randomised control trial	Young people (n=30) aged 14 to 21 years with cancer and their surrogates or families (n=30) Young people: Male (n=18); female (n=12) Race: Asian (n=2); Black (n=13); White (n=15) Condition: Leukaemia (n=14); Lymphoma (n=2); Solid tumour (n=6); Brain tumour (n=8) Education:	US	Children's National Medical Center, Washington DC	ACP enabled families to understand and honour their adolescents' wishes. Intervention dyads were more likely than controls to limit treatments. Underserved African American families were willing to participate	ACP in practice • A structured ACP process can improve communication and engagement of young people and empower them Communication • A structured ACP process can improve clinicians' communication • A structured process can help engagement of	Included Study discusses ACPfYP with the target age group. The inclusion of young people's wishes is also part of the study. Hawker et al, framework score: (35/36)

Title	Aim	Method	Participants	Country	Setting	Findings	Themes	Inclusion
			No high school diploma or were in high school (n=24) Families/surrogates: Male (n=2); female (n=28) Race: Asian (n=1); Black (n=13); White (n=15); declined (n=1) Had at least some education to college/higher education level (n=24) An income >300% federal poverty line (n=14), with only 4 families reporting an income equal to or below the federal poverty line				young people and allow them to express their wishes Relationships • A structured process provides a potential for avoiding conflict	
What Do Adolescents Want? An Exploratory Study Regarding End-of-Life Decision-making[56]	To explore whether differences exist between chronically ill and healthy adolescents with regard to their attitudes about EoL issues	Focus group with young people aged >18 and interviews with HCPs Focus group and interviews helped develop a 25-item survey,	Unspecified HCPs Young people: Healthy: Aged 14-21 (n=25) Male (n=6); female (n=19) Ethnicity: African-American (n=25); Caucasian; Hispanic	US	Children's National Medical Center, Washington DC	Teens without a chronic illness have thought about EoL issues and have opinions similar to those of chronically ill teens. Most healthy and chronically ill adolescents do want to be involved	ACP in practice • Palliative care should be integrated from the time of diagnosis • Participating in the ACP process gives young people some control over what happens to them	Included Study discusses ACPfYP with the target age group. The inclusion of young people's wishes is also part of the study

		which was then used to gather the views of young people	Chronically ill Aged 13-21 (n=25) Male (n=9); female (n=16) Ethnicity: African-American (n=19); Caucasian (n=3); Hispanic (n=3)			in shared decision-making. The EoL decision-making process should be individualised	Reliance on guidelines and recommendations to overcome barriers Communication • Tension surrounding the communication and timing of communication about ACP	Hawker et al, framework score: (33/36)
Development, Feasibility, and Acceptability of the Family/ Adolescent-Centered (FACE) Advance Care Planning Intervention for Adolescents with HIV[49]	To develop, adapt, and ensure feasibility, acceptability, and safety of the Family/ Adolescent-Centered (FACE) Advance Care Planning intervention	Two-group, randomised, controlled trial	Young people: Aged 14-21 (n=38) Male (n=15); female (n=23) Race/Ethnicity: Black/African American (n=35); White/Caucasian (n=2); American Indian/Alaskan (n=1) The majority of participants (n=22) had no high school diploma or were in high school (n=22) The majority of participants (n=35) were in permanent housing.	US	Two unspecified hospital-based outpatient clinics in Washington, D.C. and Memphis, Tennessee	Existing ACP models can be adapted for age, disease, and culture. Young people with HIV/AIDS were satisfied with an ACP approach that facilitated discussion about their EoL wishes with their families. Families acknowledged a LTC and were willing to initiate EoL conversations when their children were medically stable	ACP in practice • A structured process is more effective at engaging the target population Communication • Positive contribution of conversations about EoLC Relationships • Potential conflict between young people and family members	Included Study discusses ACPfYP with the target age group. The inclusion of young people's wishes is also part of the study Hawker et al, framework score: (35/36)

			There was a spread of different income levels The majority (n=32) were heterosexual The majority (n=36) were single						
Engaging College Undergraduates in Advance Care Planning[57]	To examine the reactions of college students following the completion of their advance care plans and then sharing these plans with friends and family	1. Students received a 90-min lecture on the importance of ACP 2. Students participated in facilitated discussion around completing their ACP 3. Students were asked to present their ACP to key family members and friends 4. Students were asked to write a 5-page reflection	3rd and 4th year undergraduate students	US	An unspecified large Midwestern university	Students reported that while completing their ACPs created many emotions, they found the experience to be valuable and facilitated conversations with family and friends about end-of-life care that may not have occurred otherwise	ACP in practice • Self-protective disengagement from some students • Worry, fear and anxiety • Positive growth (empowerment) • ACPs can foster effective communication Training/Education • Educators have a key role in starting discussions with young people about their ACP Relationships • Choosing the right family member to be their decision-maker	Included The study includes the views of young people who match my criteria. It also explores how their views can be incorporated into their own ACP alongside their family's views Hawker et al, framework score: (28/36)

							paper on the experience of completing their ACP	
How I Wish to be Remembered: The Use of an Advance Care Planning Document in Adolescent and Young Adult Populations[50]	To explore whether adolescents and young adults living with a LLC find it acceptable and helpful to have a planning document to share their wishes and thoughts regarding EOL care. If so, to learn about specific concerns adolescents and young adults feel are important to include in such a document.	Survey	20 study participants Male (n=10); female (n=10) Age range: 16-19 years (n=9) 20-28 years (n=11) Race: Caucasian (n=10) African American (n=8) Hispanic (n=2) Diagnosis: Cancer (n=10) HIV (n=10)	US	National Cancer Institute, Pediatric Oncology Branch Maryland	ACP may be appropriate and helpful for young people living with a serious illness. Future research should further validate the preferences identified by participants and explore whether an age-appropriate document can improve communication with family and staff in EoL care	ACP in practice • Young people may be less concerned with legalities and medical decision-making than treatment, memories and those they leave behind Communication • ACP may foster communication within the family	Included Discussion about the importance of including young people in their own ACP Hawker et al, framework score: (29/36)
Allowing Adolescents and Young Adults to Plan Their End-of-Life Care[53]	To assess and compare the usefulness, helpfulness, and stress associated with reviewing a previously adapted advance care planning guide, My	Ranking exercise on My Thoughts, My Wishes, My Voice and, Five Wishes	Fifty-two participants living with metastatic or recurrent cancer (n=16) or HIV infection (n=26) Aged 16-28 years	US	Pediatric Oncology Branch of the National Cancer Institute and from Georgetown University Hospital	Seriously ill young people benefit from being involved in their EoL treatment and care plans. An age-appropriate ACP can provide a sense of trust with the medical team,	ACP in practice • It is important to engage seriously ill young people in their ACP • A structured document allows greater engagement of the young people	Included Identifies the importance of engaging young people in their ACP and suggests how young people can be engaged

Title	Aim	Method	Participants	Country	Population	Findings	Themes	Notes
Thoughts, My Wishes, My Voice, in comparison with the widely used adult document Five Wishes by adolescents and young adults (AYAs) living with a serious illness			Male (46%); Female (54%) Race: African American (44%) White (33%) Hispanic/Latino (14%) Asian/Pacific Islander (2%) Biracial (4%) Other (2%)			their family, and providing some measure of control and independence for the young person Certain items are important in a specific document that allows compassionate and self-directed care, support, and comfort both before and after death. Voicing choices allows young people to reflect on their life and to make choices about what nurtures, protects, and affirms their remaining life and how they wish to be remembered into the future	and potential for compassionate and self-directed care, support and comfort both before and after death Relationships • ACP can create a sense of trust between young people, their family and the medical team	Hawker et al, framework score: (30/36)
Differences between Pediatricians and Internists in Advance Care Planning for Adolescents with Cancer[51]	To evaluate differences between paediatricians and internists in the practice of and barriers to (ACP)	Self-reported questionnaire	Paediatricians (n=227) Male (n=174); female (n=53) Age: 47.6 ± 7.9	Japan	Japanese board-certified haematology-ists	Paediatricians were less likely than internists to discuss ACP and advance directives with patients, and both paediatricians and internists tended to	ACP in practice • Specialists more likely than other HCPs to discuss ACP • Barriers to ACP tabulated - specialists	Included Discussion about the importance of including young people in their own ACP and the potential barriers and

		Various places of work:			discuss ACP and advance directives more often with patients' families	perceived more barriers than other HCPs • ACP usually discussed more with the family than young people	facilitators to engaging them Hawker et al, framework score: (29/36)
for young people with cancer		Internists (n=373) Male (n=293); females (n=80) Age: 49.1 ± 9.2					

The studies generally covered multiple themes. Only one study[44] covered a single theme. The theme(s) covered by each individual study are summarised in Table 4.

Table 4 – Summary of the themes covered by each study

Study author(s) and date of publication	Theme 1 ACP in practice	Theme 2 Communication	Theme 3 Education/training	Theme 4 Relationships
Beecham et al. (2016)[54]	√			√
Beringer and Heckford (2012)[60]	√		√	√
Billings and Holdsworth (2013)[55]	√	√	√	√
Christenson et al. (2012)[52]	√	√		
Dallas et al. (2016)[40]	√	√		
De Broca et al. (2016)[41]	√	√		√
Durall, Zurakowski and Wolfe (2012)		√	√	

Freytag and Rauscher (2017)		√		√
Garvie et al. (2012)		√		
Heckford and Beringer (2014)	√	√		
Jacobs et al. (2015)	√		√	
Lotz et al. (2015)	√			
Lotz et al. (2013)	√		√	
Lyon et al. (2017)	√			
Lyon et al. (2014)	√	√		
Lyon et al. (2013)	√	√		√
Lyon et al. (2004)	√	√		
Lyon et al. (2009)	√	√		√
Sanders and Robinson (2017)	√	√	√	√
Wiener et al. (2008)	√	√		
Wiener et al. (2012)	√			√
Yotani, Kizawa and Shintaku (2017)	√			

How ACP works in practice

How ACP works in practice and how those involved in the process want it to work was the most commonly recurring theme across the 22 studies (see Table 3). Families want their child to be engaged in their own ACP and they want their child's opinion to be heard.[40,44,53] However, participation is not always straightforward in practice. ACP can be characterised by disagreements between the family about the location of death[58]. This conflict can be compounded by a reluctance of HCPs and/or parents to engage in ACP if prognosis is poor[46,54] These issues can sometimes result in a breakdown in the relationships both within the family and between the family and HCPs.

Decisions within the ACP process are made more difficult by instability in the condition of young people. Parents generally desire care and treatment options to remain open, although decisions can be complicated by parents trying to visualise a hypothetical situation.[54] Indeed, some evidence suggests that ACP might only be started after a crisis situation or when the health of the young people suddenly deteriorates.[41,59] This means that although parents welcome ACP discussions,[54] the timing of them often occurs too late for their children to be involved. Therefore, poor timing of ACP can explain why most decisions about the future of young people rarely involve the young people themselves, despite the willingness to engage them.[54]

There are also problems about the lack of information regarding available resources to help guide ACP discussions.[58] These issues may create a barrier to the engagement

of young people by impacting on their ability to make informed decisions about treatment, care or place of death.[58] Specialist HCPs may also be more likely than other HCPs to both discuss ACP and identify barriers to the engagement of both young people and their family.[51] Little research discussed the role of different HCPs and it is not possible to draw conclusions from the evidence. Nevertheless, it may be that a more specialist team, with experience of working with young people, can help to facilitate a more positive experience for those young people around ACP.[55]

Conversely, young people were reported to be more concerned with practical issues, such as the treatment they receive and anxiety for people who are left behind after their death.[50] Yet although young people want to be involved in their own ACP,[48] they are not always given the opportunity to participate. The resulting barriers identified above suggest that a poor approach to ACP can foster self-protective disengagement from young people due to worry, fear and anxiety such discussions can produce.[57] This, in turn, results in a variety of documentation and patchy engagement of young people between different services.[39,55] In contrast, a structured ACP intervention which is targeted on positive relationships and effective communication[47,49] can facilitate young people's participation in their own ACP and contribute to the reduction of both stress and anxiety.[48] Indeed, one study concluded that ACP could take place from the time of diagnosis.[56] Such an approach can empower young people and facilitate their engagement in the ACP process regardless of perceived barriers.[48,56,57]

Communication

Poor, and the incorrect timing of, communication can cause unnecessary tension[56] and be a barrier to young people's engagement in their ACP process[44]. However, there is potential to foster effective communication about ACP within families.[43,50] The positive role of communication within the ACP process was highlighted by over a third of studies.[40,42,45,47,49,50,52,58] Indeed, high-quality communication can help young people understand the care they need and develop skills to convey their wishes.[40,48,49,51,52]

ACP should be viewed as a process which recognises the shared vision and values of different generations within the same family.[43] This approach may help to reduce death anxiety[43] and so facilitate the engagement of young people in their own ACP. HCPs should balance issues around hope for the future and respect for the individual with honest and meaningful conversations about dying.[42,52,53,58] Open communication, as part of an age-appropriate and accessible communication style, can possibly facilitate the engagement of young people.[59]

Despite their age, young people may be in the best position to discuss their own wishes, but communication can often be a barrier rather than a facilitator to their engagement in the ACP process.[42,45,47] However, the reluctance, or discomfort, of HCPs to discuss EoL matters with young people can be a barrier to their engagement in their own decision-making process.[59] This breakdown in communication too often can lead to a failure in relationships and also highlight inadequacies in training and education.[42]

Training and education

HCPs generally work at the desired pace of families and develop a flexible approach to engage them.[55] Nevertheless, access to relevant training can enhance the quality of both communication and relationships of those in the ACP process. While education for young people can be key to raising awareness of ACP,[57] access to and availability of training for HCPs was identified as a possible barrier to engaging young people in their own ACP.[42,58] For example, HCPs are not always aware of relevant policies and can sometimes be unsure of their responsibilities.[50] This can impact on their ability to engage young people in ACP discussions.[42] Poor communication skills and a unwillingness to discuss ACP with young people have also been identified as potential training needs for HCPs in order to facilitate young people's engagement.[59] Greater availability of training and guidance to support HCPs in the ACP process have started to address some of the issues identified above,[60] but these need to be ongoing to have a lasting effect.[55]

Valuable strategies, such as surveys to stimulate conversations[44] and the consideration of real-life situations,[57] can facilitate sensitive communication at opportune times. These approaches can engage young people and facilitate open and honest discussions about their wishes, fears and hopes. Similarly, inter-disciplinary HCP discussions can also help to reduce professional tensions[42] and so potentially facilitate the engagement of young people in their own ACP. However, current research indicates that this potential is not always being realised.[42]

Relationships

Assumptions about young people's understanding of ACP are sometimes based on their age. These beliefs can be a limiting factor to young people's engagement in their

own ACP.[43] Age may also contribute to tensions in relationships and discussions can sometimes be characterised by conflict because of the sensitive nature of the topic.[39,49] A structured ACP process may help reduce this likelihood for tension by providing a framework in which to discuss wishes openly and intimately[48] and give friends and family members a clear role.[43,57] Furthermore, positive relationships within families, within the multi-professional team, and between these groups can create a sense of trust between young people, their family and HCPs.[53] Developing such relationships necessitates a multi-disciplinary approach to facilitate the engagement of young people in their own ACP and involve appropriate HCPs.[41] In these situations, peer support for HCPs can enrich professional relationships[60] and so complement the role of the family in ACP.[43]

Building trusting relationships is also important in the ACP process.[54] Evidence stresses that quality relationships, which are based on mutual understanding, trust, and respect, can help recognise the wishes of each person as valuable within ACP.[41,43] Successful relationships also depend on developing effective communication and accessing relevant training[42,48,50] In short, constructive, encouraging and progressive relationships can allow ACP to flourish and facilitate the engagement of young people in their own ACP; negative, pessimistic and difficult relationships can be more of a barrier to their engagement.

Discussion

Main findings

Most studies conducted within hospitals and specialist centres identified communication as one of the main factors impacting on the success of engaging young

people in their ACP process. [40,42,48-50,52,56,58] Communication, which is clear, timely, and language which is both age- and developmentally-focused, can facilitate the engagement of young people by helping them understand, and convey, the care they want at the end-of-life.[40,48,49,51,52] This approach allows openness and honesty in all relationships in the ACP process and helps family members share wishes and values for the future.[43] Conversely, communication can often be a barrier to ACP when these issues are not recognised[44,56] and this can negatively impact the engagement of young people.

Five studies[43,48,49,53,57] established that effective relationships, characterised by this openness and honesty, as well as age-appropriate language and communication style, were central to successfully engaging young people in their ACP. Evidence also recognised ACP is a complex process, and the availability of training for HCPs is crucial to both help understand these complexities and overcome potential barriers.[57]

Additionally, training to enhance HCPs' knowledge and understanding of relevant policies and legislation, and specifically communication training, would facilitate the introduction and engagement of young people in their ACP process.[42,58] Such training can help raise awareness of ACP[57] and highlight the role of HCPs in engaging young people in ACP discussions.[42] Access to training can therefore be understood as a potential facilitator to introduce conversations about ACP, ease worries and concerns of those involved in the process and engage young people to help them communicate their EoL wishes.[61] Currently there is little evidence to show that this training for HCPs is consistent.[55,62] Consequently, availability and access to such training may act as more of a barrier to engaging young people in their own ACP.[60] Therefore, the

combined impact of strained relationships, poor communication, and restricted availability of training for HCPs presents a mixed picture of barriers ACPfYP.

Despite the barriers identified, there were also encouraging signs where young people had been engaged. For example, the alignment of practice and policy, whereby HCPs feel guidance is provided to facilitate the engagement of young people and start difficult conversations, can help young people develop skills to convey their own care treatment decisions[52,53] and develop their resilience[40]. A collaborative approach to ACPfYP would further increase the potential for compassionate and self-directed care, comfort and support both before and after death.[53]

Limitations

There are several limitations to this review. A narrative synthesis is aimed at, and supports, the combination of heterogeneous studies, but the varied nature of the studies can make it difficult to synthesise the information. Therefore, a potential for bias by over-representing one or more studies, or themes, remains a possibility. Variety in key terms and definitions in the literature used to refer to ACPfYP made searching for articles and discussing the engagement of young people in the process a challenging task. The relatively small amount of available literature also made the review additionally complicated. For example, some studies focused on parents' or HCPs' perception of the engagement of young people rather than focusing on the views of the young people themselves. Indeed, no study focused on the views and experiences of the young people within the ACP process from their own perspective. Responses from different groups within the ACP process were also sometimes combined, which made it hard to differentiate individual viewpoints. Furthermore,

some studies included in the review form a 'study cluster', where a group of studies originate from a single institution or publication.[63] This approach provides depth of understanding for the context and implementation of ACP and has been balanced with other findings in this review.[63] This depth is particularly important when researching complex interventions and establishing their effectiveness.[63] However, there may also be limitations in providing a full breadth of understanding around ACPfYP.[63] Additionally, each study has been individually assessed and only those studies which met the criteria for this synthesis have been included. As a result, previous publications they referred to have not been included if they do not meet the criteria, even if they include relevant methodological guidelines. This is an inherent limitation of qualitative assessment frameworks and represents a potential bias in the scoring of included studies. Furthermore, BH completed the scoring of the studies independently and disagreements were discussed within the research team (MoB,AF and KK). This process provided transparency and rigour but also may have created potential bias in the scoring of included studies.

While qualitative methods may be better suited to answering questions of perception, such approaches also have limitations. A key factor of the studies included in this review is the limited focus of some of the research. Nearly a quarter of studies[41,50,54,56,57] were limited to a single institution where data saturation may not be achievable. Nevertheless, the qualitative studies included in the review were useful to address the research question and their results are helpful to understand reported practice.

Furthermore, studies inevitably vary in quality. Factors which negatively impacted on the quality of research included: a lack of clarity in their aim(s);[41,52,55] a poor description of data collection and sampling methods;[52,54] incomplete data analysis;[24,39,41,50,52,53] poor consideration of ethical issues;[24,39,41-43,50,52,54,58] or a lack of generalisability and/or discussion of implications for policy and practice.[41,51,52,58] Nevertheless, only a minority of papers (n=4)[24,41,52,58] were judged either as poor or very poor in overall quality. Although this review identifies available literature, existing research is not necessarily representative of the current engagement of young people in their ACP. Consequently, more research is needed in this area. Research is also needed to investigate the experience and involvement of all participants (young people, their family and HCPs) in the ACP process.

Strengths

Despite the limitations above, the included studies and synthesis approach satisfactorily answer the review question. The synthesis was conducted primarily by one reviewer but the procedure was checked at each stage by the research team. This process reduced the potential for bias and ensured the review was conducted rigourously and is replicable. Utilising an identifiable and tested approach to the synthesis further strengthened the reliability of the results. The rigour of the literature search resulted in the comprehensive identification of relevant studies. Inclusion of all applicable studies in the synthesis allowed for a broad and full understanding of the phenomenon under review. Even with the heterogeneous nature of the studies, the findings appear similar. Most of the studies were conducted in the United States and/or in hospitals/other institutions but the overall congruence between studies gives

encouragement regarding the strength of the findings and their applicability to a variety of settings.

What this study adds

This review presents an initial picture of young people's engagement in their own ACP. As part of this, the review has identified barriers and facilitators to the engagement of young people in the complexity of their own ACP discussions. These aspects include communication, relationships and training available to healthcare professionals. These factors influence how ACP works in practice and help determine the success or failure to engage young people in the process. As far as it is known, this is the first review of its kind and highlights the importance of understanding the complex factors in ACP. The implications of these findings demonstrate consequences for practice by considering the components of the process which can either help to engage or hinder young people from participating. The conclusions can also influence policy by informing areas such as communication training and stressing the importance of multi-disciplinary teams when engaging and supporting young people's engagement in their ACP. These areas are particularly relevant for UK policy and practice because ACP documentation is not standardised and there is an unclear picture of how young people can be engaged in discussions about their care. Guidelines can be developed to recognise potential barriers and so facilitate the engagement of young people.

As a result of this review, there are several areas where future research could be conducted. The opinions and experiences of young people are not included in the current review and so the extent to which young people are able to engage in their own ACP should be explored from their own viewpoint. Similarly, there is a lack of existing

qualitative research which explores barriers and facilitators to engaging young people in their own ACP. Current research explores the engagement of young people from one or two perspectives within the process of ACP. Research is now needed to explore all groups concurrently in the ACP process: young people, their parents/carers and healthcare professionals. The role of other professionals, such as those in mental health, psychologists and case workers, also needs to be investigated. This focus will provide additional quality research to adequately meet existing gaps in knowledge.

Conclusion

With reference to the aim of this review, there can be a variety of people involved in the process of ACPfYP: parents/carers, different HCPs and, sometimes, young people themselves. The engagement of young people in their own ACP seems to be something desired by all participants involved in ACPfYP. However, this engagement appears to be inconsistent in practice. The congruence of policy and the aims of ACP are necessary to facilitate this engagement but too often, these factors do not work together effectively.

There are also various potential barriers to young people's engagement in their ACP, such as poor communication, conflict within relationships of those involved in the process and variable access to training for HCPs. Conversely, these factors can also be facilitators to engaging young people when their importance is recognised and time and resources are invested into their effective use. Specialist HCPs may be in the best position to identify these factors and take advantage of them[51], but more research is needed to confirm this.

Consequently, there is no general agreement of when it is best to introduce ACP discussions. Only two studies identified an optimal time to start ACP and both agreed that it is best introduced at diagnosis.[33] With no further investigation of the timing of ACP, this represents a gap in current knowledge.

Authorship

The review was written primarily by BH, with contributions from the rest of the research team (MoB,AF and KK). All authors have:

(i)   Made a substantial contribution to the concept or design of the work; or acquisition, analysis or interpretation of data,

(ii)   Drafted the article or revised it critically for important intellectual content,

(iii) Approved the version to be published,

(iv)   Have participated sufficiently in the work to take public responsibility for appropriate portions of the review.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Research ethics and patient consent

No ethics/research governance/data protection approvals were necessary to conduct this review.

Funding

References

1.   Hawker S, Payne S, Kerr C, et al. Appraising the evidence: reviewing disparate data systematically. *Qual Health Res* 2002; 12: 1284-1299.
2.   National Gold Standards Framework Centre. Welcome to the Gold Standards Framework. *The Gold Standards Framework*http://www.goldstandardsframework.org.uk/ (2017, accessed 12 September 2017).
3.   Seymour J, Almack K, Kennedy S. Implementing advance care planning: a qualitative study of community nurses' views and experiences. *BMC Palliat*

Care 2010; 9: 4.

4.   Reynolds J, Croft S. How to implement the Gold Standards Framework to ensure continuity of care. *Nurs Times* 2010; 106: 10-13.

5.   Lund S, Richardson A, May C. Barriers to advance care planning at the end of life: An explanatory systematic review of implementation studies. *PLoS One* 2015; 10: 1-15.

6.   Hayhoe B, Howe A, Gillick M, et al. Advance care planning under the Mental Capacity Act 2005 in primary care. *Br J Gen Pract* 2011; 61: e537-41.

7.   Montreuil M, Carnevale FA. A concept analysis of children's agency within the health literature. *J Child Heal Care* 2015; 1-9.

8.   DH. *Better Care : Better Lives Improving outcomes and experiences for children, young people and their families living with life limiting and life threatening conditions.* Bristol, 2008.

9.   Nursing Times. What is the Liverpool Care Pathway? *Nursing Times*https://www.nursingtimes.net/clinical-subjects/end-of-life-and-palliative-care/what-is-the-liverpool-care-pathway/5051586.article (2012, accessed 23 November 2016).

10.   Linda Emanuel, Michael Barry, John Stoeckle LE and E. Advance Directives for Medical Care - A Case for Greater Use. *N Engl J Med* 1991; 324: 889-895.

11.   Hynson J. Palliative care for children. *J Consum Heal Forum Aust* 2009; March: 22-23.

12.   Mitchell S, Dale J. Advance Care Planning in palliative care: A qualitative investigation into the perspective of Paediatric Intensive Care Unit staff. *Palliat Med* 2015; 29: 371-379.

13.   TfSL. Why we are here.

14.   Russell S. Advance care planning: whose agenda is it anyway? *Palliat Med* 2014; 28: 997-9.

15.   Sudore RL, Lum HD, You JJ, et al. Defining Advance Care Planning for Adults: A Consensus Definition From a Multidisciplinary Delphi Panel. *J Pain Symptom Manage* 2017; 53: 821-832.

16.   South Central NHS Trust. Guide to using the Child and Young Person's Advance Care Plan.

17.   West Midlands Paediatric Palliative Care Network NHS. *Child and Young Person's Advance Care Plan: Policy*.https://www.togetherforshortlives.org.uk/assets/0000/1485/6b_ACP_Policy_2012.pdf (2011).

18.   TfSL. Family Factsheets: Care Planning in Advance.

19.   Horridge K. Advance Care Planning: practicalities, legalities, complexities and controversies. *Arch Dis Child* 2015; 100: 380-385.

20.   Cass H. National priorities and crystal ball gazing. In: *A Palliative Care Journey: National Paediatric and Neonatal Palliative Care Conference*. 2016, pp. 1-60.

21.   Bell CJ. Understanding Quality of Life in Adolescents living with Advanced Cancer. *J Chem Inf Model* 2013; 53: 1689-1699.

22.   Bergstraesser E, Zimmermann K, Eskola K, et al. Paediatric end-of-life care needs in Switzerland: current practices, and perspectives from parents and professionals. A study protocol. *J Adv Nurs* 2015; 71: 1940-1947.

23.   NHS South East Coast Clinical Senate. *Improving Advance Care Planning in Kent, Surrey and Sussex.* 2014.

24.   Heckford E, Beringer AJ. Advance Care Planning: Challenges and Approaches

for Pediatricians. *J Palliat Med* 2014; 17: 1049-1053.

25. Mitchell S, Plunkett A, Dale J. Use of formal advance care planning documents: a national survey of UK Paediatric Intensive Care Units. *Arch Dis Child* 2014; 99: 327-30.

26. Snilstveit B, Oliver S, Vojtkova M. Narrative approaches to systematic review and synthesis of evidence for international development policy and practice. *J Dev Eff* 2012; 4: 409-429.

27. Firn J, Preston N, Walshe C. What are the views of hospital-based generalist palliative care professionals on what facilitates or hinders collaboration with in-patient specialist palliative care teams? A systematically constructed narrative synthesis. *Palliat Med* 2016; 30: 240-256.

28. Gowing A, Robinson L. Advance care planning. *InnovAiT Educ Inspir Gen Pract* 2014; 7: 363-369.

29. TfSL. Children's palliative care definitions.http://www.togetherforshortlives.org.uk/assets/0000/1638/CPC_defi nitions.pdf (2013, accessed 23 October 2015).

30. World Health Organisation. Definition and list of health professionals. *Transformative Education for Health Professionals*http://whoeducationguidelines.org/content/1-definition-and-list-health-professionals (2015, accessed 13 December 2015).

31. TfSL. *A Core Care Pathway for Children with Life-limiting and Life-threatening Conditions.* Bristol, 2013.

32. Neuberger J, Aaronovitch D, Bonser T, et al. *More Care Less Pathway: A Review of the Liverpool Care Pathway.* 2013.

33. TfSL. 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. *Epidemiology* 2011; 1-129.

34. US National Library of Medicine. MeSH Browser. *Mesh Descriptor Data*https://meshb.nlm.nih.gov/record/ui?ui=D055815 (2017, accessed 6 April 2017).

35. Liberati A, Altman DG, Tetzlaff J, et al. The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: Explanation and elaboration. *PLoS Med*; 6. Epub ahead of print 2009. DOI: 10.1371/journal.pmed.1000100.

36. Stewart LA, Clarke M, Rovers M, et al. Preferred Reporting Items for a Systematic Review and Meta-analysis of Individual Participant Data. *Jama* 2015; 313: 1657-1665.

37. Popay J, Roberts H, Sowden A, et al. Guidance on the Conduct of Narrative Synthesis in Systematic Reviews. *A Prod from ESRC Methods Program*. Epub ahead of print 2006. DOI: 10.1111/j.1523-536x.1995tb00261.x.

38. Robinson L, Dickinson C, Rousseau N, et al. A systematic review of the effectiveness of advance care planning interventions for people with cognitive impairment and dementia. *Age Ageing* 2012; 41: 263-269.

39. Beringer AJ, Heckford EJ. Was there a plan? End-of-life care for children with life-limiting conditions: a review of multi-service healthcare records. *Child Care Health Dev* 2014; 40: 176-183.

40. Dallas RH, Kimmel A, Wilkins ML, et al. Acceptability of Family-Centered Advanced Care Planning for Adolescents With HIV. *Pediatrics* 2016; 138: e20161854-e20161854.

41. de Broca A, Lutun A, Gourmel A, et al. Advance care planning and place of

death in a paediatric palliative care unit in France. *Eur J Palliat Care* 2016; 23: 16-18.

42. Durall A, Zurakowski D, Wolfe J. Barriers to Conducting Advance Care Discussions for Children With Life-Threatening Conditions. *Pediatrics* 2012; Apr 129: e975-e982.

43. Freytag J, Rauscher EA. The Importance of Intergenerational Communication in Advance Care Planning: Generational Relationships among Perceptions and Beliefs. *J Health Commun* 2017; 22: 488-496.

44. Garvie PA, He J, Wang J, et al. An exploratory survey of end-of-life attitudes, beliefs, and experiences of adolescents with HIV/AIDS and their families. *J Pain Symptom Manage* 2012; 44: 373-385.e29.

45. Lyon ME, Jacobs S, Briggs L, et al. Family-Centered Advance Care Planning for Teens With Cancer. *JAMA Pediatr* 2013; 167: 460.

46. Lotz JD, Jox RJ, Borasio GD, et al. Pediatric Advance Care Planning: A Systematic Review. *Pediatrics* 2013; 131: e873-e880.

47. Lyon ME, D'Angelo LJ, Dallas RH, et al. A randomized clinical trial of adolescents with HIV/AIDS: pediatric advance care planning. *AIDS Care - Psychol Socio-Medical Asp AIDS/HIV* 2017; 29: 1287-1296.

48. Lyon ME, Jacobs S, Briggs L, et al. A Longitudinal, Randomized, Controlled Trial of Advance Care Planning for Teens With Cancer: Anxiety, Depression, Quality of Life, Advance Directives, Spirituality. *J Adolesc Heal* 2014; 54: 710-717.

49. Lyon ME, Garvie PA, Briggs L, et al. Development, feasibility and acceptibility of the Family/Adolescent-Centred (FACE) Advance Care Planning Intervention for Adolescents with HIV. *J Palliat Med* 2009; 12: 363-372.

50. Wiener L, Ballard E, Brennan T, et al. How I Wish to be Remembered: The Use of an Advance Care Planning Document in Adolescent and Young Adult Populations. *J Palliat Med* 2008; 11: 1309-1313.

51. Yotani N, Kizawa Y, Shintaku H. Differences between Pediatricians and Internists in Advance Care Planning for Adolescents with Cancer. *J Pediatr* 2017; 182: 356-362.

52. Christenson K, Lybrand SA, Hubbard CR, et al. Including the Perspective of the Adolescent in Palliative Care Preferences. *J Pediatr Heal Care* 2010; 24: 286-291.

53. Wiener L, Zadeh S, Battles H, et al. Allowing adolescents and young adults to plan their end-of-life care. *Paediatrics* 2012; 130: 897-905.

54. Beecham E, Oostendorp L, Crocker J, et al. Keeping all options open: Parents' approaches to advance care planning. *Heal Expect* 2016; 1-10.

55. Billings J, Holdsworth L. *Service Evaluation of an Advance Care Plan Tool*.http://www.kent.ac.uk/chss/docs/advance-care-plan-tool-report.pdf (2013).

56. Lyon ME, McCabe MA, Patel KM, et al. What do adolescents want? An exploratory study regarding end-of-life decision-making. *J Adolesc Heal* 2004; 35: 529.e1-529.e6.

57. Sanders S, Robinson EL. Engaging College Undergraduates in Advance Care Planning. *OMEGA - J Death Dying* 2017; 74: 329-344.

58. Jacobs S, Perez J, Cheng YI, et al. Adolescent End of Life Preferences and Congruence With Their Parents' Preferences: Results of a Survey of Adolescents With Cancer. *Pediatr Blood Cancer* 2015; 62: 710-714.

59. Lotz JD, Jox RJ, Borasio GD, et al. Pediatric advance care planning from the

perspective of health care professionals: A qualitative interview study. *Palliat Med* 2015; 29: 212-222.

60. Beringer AJ, Heckford EJ. Was there a plan? End-of-life care for children with life-limiting conditions: A review of multi-service healthcare records. *Child Care Health Dev* 2012; 40: 176-183.

61. Durall A, Zurakowski D, Wolfe J. Barriers to Conducting Advance Care Discussions for Children With Life-Threatening Conditions. *Pediatrics* 2012; Apr 129: e975--e982.

62. Lyon ME, Williams PL, Woods ER, et al. Do-Not-Resuscitate Orders and/or Hospice Care, Psychological Health, and Quality of Life among Children/Adolescents with Acquired Immune Deficiency Syndrome. *J Palliat Med* 2008; 11: 459-469.

63. Booth A, Harris J, Croot E, et al. 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* 2013; 13: 118.

Appendix 1 – Assessment framework [1]

Author and title:

Date:

	Good	Fair	Poor	Very poor	Comment
1. Abstract and title					
2. Introduction and aims					
3. Method and data					
4. Sampling					
5. Data analysis					
6. Ethics and bias					
7. Findings/results					

8. Transferability/generalisability					
9. Implications					

Total	

Appendix 2 – Assessment framework[1]

---

1. Abstract and title: Did they provide a clear description of the study?

Good   Structured abstract with full information and clear title
Fair   Abstract with most of the information
Poor   Inadequate abstract
Very poor   No abstract

---

2. Introduction and aims: Was there a good background and clear statement of the aims of the research?

Good   Full but concise background to discussion/study containing up-to- date
                literature review and highlighting gaps in knowledge
            Clear statement of aim AND objectives including research questions
Fair   Some background and literature review
            Research questions outlined
Poor   Some background but no aim/objectives/questions, OR Aims/objectives but
                inadequate background
Very poor   No mention of aims/objectives
            No background or literature review

---

3. Method and data: Is the method appropriate and clearly explained?

Good   Method is appropriate and described clearly (e.g., questionnaires included)
            Clear details of the data collection and recording
Fair   Method appropriate, description could be better
            Data described
Poor   Questionable whether method is appropriate
            Method described inadequately
            Little description of data
Very poor   No mention of method AND/OR
            Method inappropriate AND/OR
            No details of data.

---

4. Sampling: Was the sampling strategy appropriate to address the aims?

Good

	Details (age/gender/race/context) of who was studied and how they were recruited
	Why this group was targeted
Fair	The sample size was justified for the study
	Response rates shown and explained
Poor	Sample size justified
Very poor	Most information given, but some missing
	Sampling mentioned but few descriptive details
	No details of sample

5. Data analysis: Was the description of the data analysis sufficiently rigorous?

Good	Clear description of how analysis was done
	Qualitative studies: Description of how themes derived/respondent validation or triangulation
	Quantitative studies: Reasons for tests selected hypothesis driven/numbers add up/statistical significance discussed
Fair	Qualitative: Descriptive discussion of analysis.
	Quantitative
Poor	Minimal details about analysis
Very poor	No discussion of analysis

6. Ethics and bias: Have ethical issues been addressed, and what has necessary ethical approval gained? Has the relationship between researchers and participants been adequately considered?

Good	Ethics: Where necessary issues of confidentiality, sensitivity, and consent were addressed
Fair	Bias: Researcher was reflexive and/or aware of own bias
Poor	Lip service was paid to above (i.e., these issues were acknowledged)
Very poor	Brief mention of issues
	No mention of issues

7. Results: Is there a clear statement of the findings?

Good	Findings explicit, easy to understand, and in logical progression
	Tables, if present, are explained in text
	Results relate directly to aims
	Sufficient data are presented to support findings
Fair	Findings mentioned but more explanation could be given
	Data presented relate directly to results
Poor	Findings presented haphazardly, not explained, and do not progress logically
Very poor	from results
	Findings not mentioned or do not relate to aim

8. Transferability or generalizability: Are the findings of this study transferable (generalizable) to a wider population?

Good	Context and setting of the study is described sufficiently to allow comparison with other contexts and settings, plus high score in Question 4 (sampling)
Fair	Some context and setting described, but more needed to replicate or compare the study with others, PLUS fair score or higher in Question 4
Poor	Minimal description of context/setting
Very poor	No description of context/setting

---

9. Implications and usefulness: How important are these findings to policy and practice?

Good	Contributes something new and/or different in terms of understanding/insight or perspective
	Suggests ideas for further research
	Suggests implications for policy and/or practice
Fair	Two of the above (state what is missing in comments)
Poor	Only one of the above
Very poor	None of the above