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Understanding care when cure is not likely for young adults who face cancer: a realist analysis of data from patients, families and healthcare professionals.

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Author Contributions

All authors were involved in study design, review of results and review of this manuscript. Interviews and analysis were conducted by CK and NN: aided by data analysis meetings with LJ, SP, FG, and GW. Workshops were attended by CK, NN, MF, LCS, FG, JW: KB and ABH attended 1 workshop and led another. Participant recruitment was assisted by MF, KB, SH, AH, LCS, JW.

Data sharing statement

No data from interviews with patients, families and health care professionals is available in the public domain given the potential for identification of sensitive information.

Additional submissions

Figures	Attached
Original protocol	Attached
Methodology protocol and Interview questions	Attached
SRQR checklist	Attached

Abstract

Objectives: To understand the experiences of young adults with cancer for whom cure is not likely, in particular what may be specific for people aged 16-40 years and how this might affect care.

Design: We used data from multiple sources (semi-structured interviews with people with cancer, nominated family members and healthcare professionals, and workshops) informed by a preliminary programme theory: realist analysis of data within these themes enabled revision of our theory. A realist logic of analysis explored contexts and mechanisms affecting outcomes of care.

Setting: Three cancer centres and associated palliative care services across England.

Participants: We aimed for a purposive sample of 45 people with cancer from two groups: those aged 16-24 years for whom there may be specialist cancer centres and those 25-40 years cared for through general adult services; each could nominate for interview one family member and one healthcare professional. We interviewed three people aged 16-24 years and 30 people 25-40 years diagnosed with cancer (carcinomas; blood cancers; sarcoma; central nervous system tumours) with an estimated prognosis of less than 12 months along with nominated family carers and healthcare professionals. Nineteen bereaved family members and 47 healthcare professionals participated in workshops.

Results: Data were available from 69 interviews (33 people with cancer, 14 family carers, 22 healthcare professionals) and six workshops. Qualitative analysis revealed seven key themes: loss of control; maintenance of normal life; continuity of care; support for professionals; support for families; importance of language chosen by professionals; financial concerns.

Conclusions: Current end-of-life care for young adults with cancer and their families does not fully meet needs and expectations. We identified challenges that are specific to those aged 16-40 years. The burden that care delivery imposes on healthcare professionals must be recognised. These findings can inform recommendations for measures to be incorporated into services.

Strengths and limitations of this study

1. There is a lack of empirical research, policy and expert practice to inform delivery of optimal care for young adults, and support of their families, when cure of their cancer is not likely.
2. Data were therefore collected by interviewing young adults under 40 years of age with incurable cancer, their nominated family carers and healthcare professionals.
3. Further primary data were obtained in workshops with bereaved family members and professionals involved in end-of-life care for young adults.
4. People with blood cancers and those aged between 16-24 years were difficult to recruit and may have unrecognised specific needs.
5. Analysis of this unique data set has highlighted specific challenges for young adults, their families and healthcare professionals in the delivery of end-of-life care.

Background

Cancer in young adults under 40 years is notable because it comprises a wide range of malignancies, has specific challenges to improving both length and quality of life but is relatively uncommon.(1) One quarter of all deaths in the United Kingdom in people aged 16-40 years are from cancer.(2) In Europe there are over 27,000 deaths per year in this age group.(3) Despite increasing empirical evidence of the specific needs of young adults in specialist cancer care, there is little evidence about their experiences at the end-of-life.(4, 5)

Boundaries between curative and palliative cancer treatments are often blurred as decisions may be influenced by cancer type, age and family circumstances as well as the experience and skills of healthcare professionals (HCPs). Avoidance and delaying of discussions about end-of-life decisions are common, often affecting the quality of care.(6) Professionals consistently acknowledge the challenges of managing end-of-life care for people in this age group, which may have commonalities with and, importantly, differences from those of people with cancer at other ages as death approaches. (7-9)

To develop our knowledge of end-of-life care in adolescents and young adults aged 16-40 years (referred to in this paper as 'young adults') with cancer, we sought to collect data directly from young adults who were facing a poor prognosis, their families and HCPs involved in their care. To gain a deep understanding of the contexts that may be specific to this age group, we chose to explore our data using a realist evaluation approach.(10) A realist evaluation approach focuses on explanations, taking account of contexts and mechanisms that may affect outcomes. It addresses questions about what works for whom, in what circumstances and in what respects, and how?

Consistent with the realist method, we began our research with a preliminary programme theory informed by expert opinion within our research team which was led by clinical academic specialists in the care of young adults with cancer. Our thinking was also informed by a narrative review of the existing literature, Phase i of our study, previously reported.(4)

Our preliminary programme theory was:

'That there are specific differences in experiences of and preferences for care towards the end-of-life for those with cancer aged 16-24 and 25-40 years compared to those who are older. Life-threatening illness in the young is untimely, it disrupts expected biographies, and maintaining a sense of control and normality in everyday life may be important. The role of close family members is complex and integral to the experiences of the person with cancer.'

We used this theory to develop topics for use in semi-structured interviews with young adults with cancer, family members and HCPs, and to underpin scenarios used in workshop discussions with HCPs and bereaved family members.

In this paper, we describe data arising from these interviews and workshops. We used our data analysis to further explore and develop the context, mechanisms and outcomes that may explain parts of our preliminary programme theory. We develop a revised programme theory that can be used to underpin recommendations for policy and practice and inform future research.

Methods

A multi-method realist study was undertaken (Figure 1). A realist evaluation approach was used as we wanted to explain and understand contextual influences on the experiences of and preferences for care towards the end-of-life for those with cancer aged 16-24 and 25-40. Here we report on Phases ii-iv, using RAMESES standards for reporting realist evaluations.(11) Phase v will be reported separately.

Recruitment and participants

We aimed to recruit a purposive sample of 45 people aged 16-40 with cancer, in two cohorts with an expected prognosis of less than one year, across four cancer groups: carcinomas; leukaemia and lymphoma; bone and soft tissue sarcoma; and central nervous system (CNS) tumours. In cohort 1 we planned to recruit 15 participants aged 16-24 years, to be interviewed at two time points; recruitment began via a national cohort study investigating whether specialist cancer services add value (www.brightlightstudy.com) and was later extended, due to poor recruitment, to include five principal treatment centres and a hospice for young adults. Cohort 2 was recruited from three specialist cancer services and three hospices in England and consisted of a purposive sample of 30 participants between the ages of 16-40. All cohort 2 participants were invited to nominate a family member and HCP involved in their care for interview.

Data collection

Semi-structured interviews

All participants took part in a semi-structured interview at a single time point using a topic guide. Cohort 1 participants were invited to take part in a later second interview.(10) We explored medical, social, communication and decision-making experiences for people with cancer and their families. We asked HCPs to reflect on the care of the person with cancer and their practice with those approaching the end-of-life. Interviews were audio-recorded and transcribed verbatim.

Workshops

We held workshops in London, Southampton and Leeds. Discussions were audio-recorded and field notes taken.

1. Healthcare professionals

Three workshops involved HCPs working in both hospital and community settings who were recruited by the sites. Scenarios were developed from information from the interviews (Table 1) and were presented to participants who were asked to discuss issues which arose.

2. Bereaved relatives

We held three workshops with bereaved relatives who were invited to take part by bereavement services in participating hospices. The workshops involved open discussions and sought to collect information that had not emerged previously in the interviews, particularly concerning the last days of life.

Table 1. Scenarios used in health care professionals' workshop

Scenario 1: 16-40 year old patient with haematological malignancy	**Scenario 2: 25-40 year old patient with oncological malignancy**
Mannu, 19, diagnosed with Hodgkin's disease in December 2013. Between December and June treated with curative intent. Relapsed June 2014 – no sibling bone marrow donor available – deteriorated before one could be found. Social Science student – sporty. University not local. Friends all at University. School friends all over country also at University. Keeps in touch with friends via Facebook. Home Returned to live with Mum, Dad and sister aged 12. Grandparents supportive – all aware of diagnosis and prognosis. Sikh faith. Supportive in background. Home is a three bed semi with a bathroom upstairs and downstairs toilet. November 2014 Inpatient. Deteriorating – wants to be at home. Unable to do stairs therefore need to make adaptations. • Symptoms – shortness of breath, cough and fatigue.	Helen, 38, diagnosed with colon cancer in May 2014. Helen lives with her partner and their 18 month old baby. Soon after diagnoses she had surgery for a stoma fitting and was diagnosed with liver metastases a few weeks after. Social She has support from her parents, brother and her partner's parents. She is currently on sick leave and misses friends from the office. They have reduced income due to her being on maternity leave before her diagnosis, although she has critical Illness Policy which will pay off their mortgage and so this is reassuring for her. Home She lives an hour's drive from her parents in a duplex house with stairs. She is getting more symptomatic and experiencing fatigue. Partner is concerned about coping with a young child and partner as she deteriorates. September 2014 Helen has lost weight and is aware that she is getting weaker and has difficulty picking

• Care – family keen to do. • Discharge home with Community Palliative Care Team input. • Contact with charities - Willow Foundation, CLIC Sargent <u>December 2014</u> Increased fatigue. Treated with radiotherapy to chest. Cough and fatigue. <u>January 2015</u> Further deterioration. Bed bound. Home oxygen. Anticipatory medications.	up/carrying her child. She is currently on a 24/7 syringe driver and the District Nurse visits daily. Referral to hospice palliative care has been made but she has not yet been in contact. Helen is referred for a clinical trial as still relatively well and no conventional treatment options. <u>December 2014</u> Chemotherapy stopped as disease not responding - parents devastated. Parents not able to access psychological support as they live 'out of the area' Advanced care planning with clinical nurse specialist causes tension as parents do not wish Helen to be 'not for resuscitation' <u>February 2015</u> House requires adaptations due to her physical condition. Increasingly housebound due to steps and steep hill Partner feels he can no longer cope as Helen's condition deteriorates further.

Data Analysis

Data were entered into a qualitative analysis software programme, NVivo 10 to facilitate analysis. (12) A realist evaluation approach enabled us to identify and understand (a) the outcomes for young people receiving care; (b) when these outcomes were likely to occur (the contexts); and (c) why (the mechanism).(10) Our analysis was multi-staged:

- *Stage One - identification of emergent themes.* Charmaz's approach was used.(13) Initial codes (summary of what participants were describing) were open and inductive from the data using verbatim quotes or researcher-generated codes to inform a conceptual framework. We then developed categories by grouping similar codes. The categories were discussed and further refined into themes.
- *Stage Two - realist logic of analysis.* This stage was undertaken as we wanted to develop findings that had a clear warrant for transferability. In other words, by reanalysing our themes, using a realist logic of analysis, we would be able to identify the commonly occurring mechanisms within this population group that caused the outcome patterns we had found. Reinterpretation of the themes was undertaken by CK and NN aided by data analysis meetings with LJ, SP, FG, and GW. To assist the reinterpretation process, we developed 'mini' programme theories that explained the care pathways and experiences of the patients, family members and HCPs. For each of these mini programme theories we re-analysed the data that we drew on to

develop each theme to build context-mechanisms-outcome (CMO) configurations - i.e. develop realist causal explanations of outcomes that occurred within different contexts (e.g. social rules and cultural systems). Workshop data were analysed in the same two-step manner and used to confirm, refute or refine the CMO configurations within the 'mini' programme theories.

Ethical review

The study was approved by Central London Research Ethics Committee (Reference: 13/LO/1098) and informed consent was sought from all participants at the time of participation.

Patient involvement

We sought the views of people with cancer on study design and written information including patient information sheets through the Cancer Partnership Research Group of the Surrey, West Sussex and Hampshire Cancer Network and the National Cancer Research Network Consumer Group. An independent steering committee, which included a bereaved parent of a young adult, provided advice and oversight on study conduct.

Results

Table 2 summarises the participants by cohort. A total of 69 interviews were conducted (33 people with cancer, 14 family members, 22 HCPs); 19 bereaved family members and 47 HCPs took part across six workshops.

Table 2 Participant Details

Cohort 1 N=30		Cohort 1 N=30	Cohort 2 N=3
Gender	Male	11	3
	Female	19	0
Age	Median (range) years	32 (16-39)	
Ethnicity	White British	19	3
	Any other White background	4	
	Asian/Asian British/Black/African/Caribbean/Black British	7	
Cancer type	Carcinoma	18	1
	Sarcoma	6	2
	Blood cancer	2	
	Other (incl. melanoma/CNS)	4	
Education/Working	Working Part Time	2	

	Working Full Time	2	
	Sick Leave	9	2
	Sick leave from education	2	1
	Not Working/Early retirement	14	
Nominated, interviewed family or other	Husband/Wife/Partner	5	
	Parent/sibling	8	
Nominated, interviewed healthcare professionals	Clinical nurse specialist	13	
	General Practitioner	2	
	Hospital doctor	4	
	Allied Health Professional	3	
	Patient did not nominate	5	
	Healthcare Professional declined participation	3	

Cohort 1 N=30		
Gender	Male	11
	Female	19
Median age (range)	Median = 32, Mean =31, range – 16-39 years	
Ethnicity	White British	19
	Any other White background	4
	Asian/Asian British/Black/African/Caribbean/Black British	7
Cancer type	Carcinoma	18
	Sarcoma	6
	Blood cancer	2
	Other (incl. melanoma/CNS)	4
Education/Working		
	Working Part Time	2
	Working Full Time	2
	Sick Leave	9
	Sick leave from education	2
	Not Working/Early retirement	14
Nominated, interviewed family or other	Husband/Wife/Partner	5
	Parent/sibling	8
Nominated, interviewed healthcare professionals	Clinical nurse specialist	13
	General Practitioner	2
	Hospital doctor	4
	Allied Health Professional	3
	Patient did not nominate	5
	Healthcare Professional declined participation	3

Cohort 2 N=3	All male, age range 22-26, all White British, 2 sarcoma and 1 carcinoma, 2 sick leave, 1 sick leave from education	

The results are presented in three sections:

1. Our thematic analysis of qualitative participant data.
2. Realistic logic of analysis reporting context, mechanisms and outcome (CMO) configurations developed from re-analyses of the themes.
3. The connections and links between contexts, mechanisms and outcomes as leading to the revision of our programme theory.

Section 1 Thematic analysis

Seven key themes emerged each of which is accompanied by one or more illustrative verbatim section of texts from our data.

Loss of Control

As illness progressed and young adults with cancer became more debilitated, they often felt a loss of control over how they lived their lives. This was a shift from independence to a growing dependence on others for physical, emotional, practical or financial support provided by family, friends, HCPs or the wider state. The future became unpredictable and planning was difficult. Maintaining a sense of control and continuing to take part in activities, albeit compromised, was important:

My independence. For me, being able to do things on my own is definitely something that I miss, without - being carefree, I can't be carefree, I can't just go out and have, get drunk with friends any more. I can't go out for a long night and dress up in heels and get bashed about, because I have a port in, I've got cancer, you know, I have to go and sit down at a bar, have a non-alcoholic cocktail. It doesn't mean I can't socialise and have a good time with them, I still do. But I'm uncomfortable when I dress up now, whereas before I had the figure and went to the gym and felt more comfortable in myself. (Cohort 2 – Patient 20)

Maintenance of Normal Life

Participants all desired to continue, as far as possible, living a 'normal life' e.g. working, taking part in activities, looking after their children. Normality provided reassurance and a sense of control but it could also be a defensive response and a shield of denial about the realities of dying from cancer. As the disease progressed the sense of 'what was normal' needed to be reframed and adjusted:

I'm at probably the worst stage I've ever been with this illness, obviously because it's more advanced. Yet people are just saying, "You're looking great." And when I look in the mirror, I don't feel like I've got cancer. I don't feel like – obviously I do because I know that I do, but I don't feel any different to how I used to feel. Obviously yes you've got a few aches and pains and stuff, but you think like, when you hear someone's dying of cancer, you think that person will feel like they are. But like I know that I am, but I don't feel like I am, because it's quite a disconnect of like how – you know, like when you're feeling alright and you're going round doing stuff, and you're just doing stuff like everyone else, you just kind of forget. You go to work and you just have the same sort of, do the same things you were doing before you had cancer. You just forget, I forget sometimes (Cohort 2 – Patient 19)

Continuity of Care

Young adults valued being known by the HCPs involved in their care and preferred a joined-up care pathway between them, the HCPs and other health services. This relied on maintaining continuity of communication and information between HCPs, services and themselves with a shared knowledge of the care plan. They generally preferred to be seen by the same HCPs as they felt they could build rapport and feel known as a person. When they moved between services e.g. from oncology to palliative care or from hospital to hospice, they wanted this to be a joined-up seamless shift:

So we went into this initial meeting and [1st tumour CNS], who is the CNS, was there. And Dr [Consultant] was the one that kept us waiting. And it was said at that point, "[1st tumour CNS] will be your CNS, presumably key worker, throughout this process, she will be at every one of your appointments when you come to clinic." And I was like, great, and he gave me her number and a pack and, you know, I felt quite supported by that. … I understand not being able to the same nurse every time, that's not possible, but like if you had a team that were allocated a certain number of patients – because they just, they don't know you. And I've noticed that across the course of having another lot, you know, and I've really – I've kind of got to know a lot of them because I've been there, you know, over the course of a year. But, you know, it is at the beginning, it's someone different every week. And they don't know anything about you. And I went in expecting them to have read my notes, know what kind of cancer it was, know, you know, some of my background, and totally naively – they – and I think it's unfair to them, they are there just to administer medication (Cohort 2 – Patient 29)

Professionals Need Support

Professionals in either cancer or palliative care settings tended to have greater experience of caring for older adults. They had less experience providing end-of-life care to those aged 16-40 and fewer 'tools' or strategies to offer this younger population. Professionals found caring for young adults as they deteriorated both challenging and burdensome. The availability, accessibility and use of support for HCPs was variable and ranged from peer to professional support with a perception that experienced senior doctors were less likely to be in need. In contrast, nurses were perceived to be more likely to require and/or seek out support:

But there's always been this sort of demarcation that when they come to the – come to, "They're now incurable," they go somewhere else. And that 'somewhere else' is always nebulous. 'Someone else' looks after them 'somewhere else.' Do you know what I mean? … "Oh they go over there now." As I said earlier, the palliative team will look after them. And I don't think any of us [Oncology CNS] have ever really gone to see what the palliative team do or see how much input they have. And is that a, is that a lack of professionalism or is that a survival mechanism for ourselves? And I have a feeling it's the latter. I have a feeling that it's very much a survival mechanism for ourselves because then we can just close that bit off and we can get on over here. And we'd like to know how they are, but we don't have to be the one that tells them. (Cohort 2 – HCP 16)

Families Need Support

Families provided multiple types of support (practical, physical, emotional, financial) to the person with cancer to complement or supplement professional care:

Well I do as much for her as I can and I go out and do all her shopping. And if her husband is not around to pick the kids up from school, myself and my other daughter, we sort of take care of the kids. And also we've been taking them out as well because – and that upsets [name] more especially during the school holidays last week that they couldn't go anywhere. And she started saying, "I'm not a good mum." (Cohort 2 – Family 22)

The impending decline and death of a young family member was usually unanticipated and a situation that families have rarely experienced before. Family members generally had fewer appropriate skills to care for the person as their cancer progressed. Families expressed a wish for some form of access to information or training to care for their loved one appropriately. Looking back, bereaved families commented that their skills to deliver care at the end-of-life were limited and they would have liked access to some basic training and emotional support.

Language

The use of language by HCPs to describe an approach to care may not convey the same meaning to young adults with cancer and their families. For example, words such as hospice conjured up particular scenarios and carried ambiguity about the imminence of the end-of-life; such terms were often left unexplained, causing distress:

I do remember him [Consultant] saying, I can't really remember the conversation massively, but I do remember him keep saying, "Tumour, there's a tumour." And then I literally did have to say, "Hang on a minute, do you mean cancer?" and he said, "Yes, we've got to run more tests and this, that and the other, but yes." But that's the only thing I remember really about it, if you know what I mean. (Cohort 2 – Patient 14)

Financial Concerns

There were few participants for whom finance was not a concern. For those who were younger and still in education or training the burden tended to fall on their families. For those who were working, with loans, mortgages or dependents, the impact of cancer compromised their ability to support themselves and their families. Concerns were expressed about changes in lifestyle whereby the basics were prioritised. There was some confusion around entitlement to benefits or equivalent sources of financial support and limited access to tailored financial advice or guidance:

But you could do with somebody saying to you, in the first place, "You need somebody to help you to do this," you know what I mean, you need somebody who can guide you through the system. And I think the same applied with [name]. He'd think, "Oh well I've just got to fill this form in and I've got..." but actually filling those forms in is a damned hard job. (Cohort 2 – Family 23)

You haven't asked to be in that position [dying from cancer]. So I shouldn't have to go to work and think, 'Well I'll do a monotonous job just to pay the bills to only live another few months.' If I've only got a few more months to live, I'd rather spend it with my family, you know, having the time with them. (Cohort 2 – Patient 6)

Section 2 Realist explanations of our themes presented in the form of Context-Mechanisms-Outcome (CMO) configurations

We re-analysed our emergent themes using a realist logic of analysis. We attempted to identify mechanisms (generative causal processes) that are activated in the contexts we had found within the themes we uncovered. Our interview data were purely qualitative and so likely to be limited in the range of relevant data needed to build CMO configurations. To supplement these data, we deliberately drew on the extensive content expertise of the project team, workshops and where relevant, existing theories on needs of people living with cancer

Details summarising the CMO configurations are presented in Table 3.

Table 3 CMO configurations, illustrative quote and summary of our interpretations

Context-Mechanism- Outcome Configuration	Quote	Related theme
CMO 1 The diagnosis of cancer (context), changed the perception of control (mechanism) in young adults to cause distress, frustration and anger (outcomes).	*… like the feeling that I've got control over it, like complete control. For me that's extremely important. As soon as I lose that, I think I'd really struggle. And I need to, yes, feel as though I'm in the driving seat more or less. (Cohort 1 – participant 1)*	From the Loss of Control theme: The unexpected diagnosis of cancer disrupted everyday life and young adults often had to relinquish control and permit others to manage aspects of their life. The feeling of 'loss of control' was experienced throughout the diagnostic and treatment phases and seemed to increase when cure was not likely as participants experienced a loss of their anticipated future.
CMO 2 In the context of disease progression (context), young adults continued with normal activities as a coping strategy that offered distraction (mechanism) leading to a feeling of some kind of 'normality' (outcome). A poor prognosis and physical decline compromised the maintenance of a 'normal life'.	*"Yes but we're not going to do that" he [son] said, "We're just going to carry on as normal." And I thought actually he's right because carrying on normal makes it, it does make it more real. And more memorable … yes we do some lovely things, but it's just trying to keep everything as normal as possible really and just make the most of that time [Cohort 2 – family member – son did not participate]*	From the Maintenance of Normal Life theme: Young adults wanted to live as normal a life for as long as possible. Young adults and their families adjusted to a new normality, to accommodate the changes their disease progression created.
CMO 3 When there was trust between HCPs and a young adult (context), it was easier to introduce change or a new service (outcome) because a sense of abandonment (mechanism) or apprehension	*I think continuity is one thing that I'd put on a pedestal as being the most important, as a patient. It's horrible seeing different people and having to tell your story over and over and over again. Yet when you see somebody you know, and they know*	From the Continuity of Care theme: Young adults reported a high expectation and preference for continuity of care within and between services, which for them meant seeing the same HCPs whenever possible:

(mechanism) was less likely to occur. In contexts where continuity was provided (i.e. seeing the same HCP) trust developed (outcome) because of a sense of being known (mechanism).	*your story, they know whether you're well or you're not well. They know how your psychology works a little bit. So they know how to present things to you. That makes a huge difference to how you trust them, what your relationship is like and how you respond to them [HCP's] (Cohort 2 – Participant 7).*	
CMO 4 When a young person is dying because of cancer (context), HCPs find it challenging to talk about a poor prognosis or a shift in goal of treatment (outcome) because of their personal emotional discomfort of such discussions (mechanism).	*I find some of the younger patients it feels very unfair and I do reflect a lot on my own mortality and how I would cope (Cohort 2 – Participant 1 HCP)* *And it's a very stark contrast that [support] seems to be important for nurses but it's not seen to be important for doctors … as an individual, you don't talk because nobody wants you to talk about it, because you're the strong leader … Some doctors will just completely divorce themselves from it and will not engage in any shape or way with their patients … But I think you just sort of, you potentially just end up with, you know, increasingly tired and burnt out and disengaged doctors (Consultant Oncologist, HCP workshop 1)*	From the Professionals Need Support theme: The loss of a 'life partially lived' can be difficult for professionals, who feel a greater burden of sadness when young adults die HCP workshop participants felt support for them was often reactive rather than proactive with nurses more likely to receive support than doctors. A further distinction was recognised between junior and senior doctors:
CMO 5 When the way services are delivered for young adults does not fully recognise the additional needs of the family or care-givers (context), this leads to	*It is quite upsetting because, we actually felt abandoned, I felt abandoned.* *[Facilitator: By?]* *By just the whole system really. It was just, if you*	From the Families Need Support theme: Families often provided informal care for young adults within the home. Whilst they wanted to support their child or partner, caring created a

them feeling marginalised (mechanism) resulting in feelings such as abandonment and distress (outcomes).	*didn't ask, you wouldn't know (Family workshop 2- bereaved husband)* *And she [24 yr. old daughter] sort of became more and more sleepy and distant from us. But nobody would say to me, "This is what to look for. When she dies, this is what's going to happen. (Family workshop 3 – bereaved mother)*	further burden. Families felt insufficiently supported in this role.
CMO 6 When emotive language is used in palliative and end-of-life care (context), misunderstandings (mechanisms) can easily occur, leading to a range of different outcomes from encouraging hope through to despair (outcomes).	*So when my breast care nurses referred me to a hospice, I was like "Oh my god, that's horrendous, I don't want to do it". But, you know, it's been one of the best ever things. And I kept putting it off and saying, "I'm not ready for it, I'm not ready for it." And she [CNS] went, "Look, if you just make contact, then when you do need them, you can tap into them and they're quite good at financial advice". So I said, "Okay right let's do it". And actually they've been fantastic. Actually from just the level of sorting things out. (Cohort 2 – participant 19).*	From the Language theme: One example was the use of the term 'hospice'. When this was first raised with young adults their initial reaction was one of rejection as hospices were where older people went to die and young adults did not believe that they were at this stage:
CMO 7 Few young adults have thoughts about long term financial planning as they did not anticipate serious illness (context). This can lead to individuals and	*You haven't asked to be in that position. So I shouldn't have to go to work and think, 'Well I'll do a monotonous job just to pay the bills to only live another few months.' If I've only got a few more*	From Financial Concerns theme: Financial concerns and insecurity are almost always expressed by young adults with cancer.

families facing financial precariousness (mechanism). Access to tailored advice, whilst it may not solve financial concerns, may provide individuals and their families a range of 'tools' to better cope with their financial situation (outcome).	*months to live, I'd rather spend it with my family, you know, having the time with them. (Cohort 2 – Patient 6)* *But, yes, I think that's the crappest thing, it's not having – if you are single and I don't have a rich family, you know, yes it's just the whole worry of like affording things and knowing what kind of life you're going to end up with if you give up work, especially when you've been used to a different kind of life. (Cohort 2 – Patient 19).*	

CMO: context, mechanism and outcome

Section 3 Revision of preliminary programme theory

Our re-analyses of the data has enabled us to confirm, further develop and refine aspects of our preliminary programme theory – namely control, normality and family support. We were also able to add to our preliminary programme theory the concepts of continuity, professional support, language and financial support. Below we summarise important aspects of our better refined programme theory.

Age specific issues

We now understand that *for those aged 16-40 there are specific differences between the end-of-life care experience and preferences.* However, *rather than being wholly defined by age, the stages in a young person's life course may be a better way to approach, understand and support these differences.*

Maintenance of control and sense of normality

Our data underpin these concepts within our preliminary programme theory. We have learned that *young adults with cancer need support to put strategies in place to retain control and live as normally as possible whist providing a space to discuss and plan for their shortened future.*

Families of younger people with cancer

We found that *the family often are not appropriately equipped to provide the level of care and support that they want to provide during the last year of life of the young adult with cancer and lack the means to be 'skilled-up' for this role.*

Healthcare professionals

We found that *healthcare professionals lack age (16-40-year-old) life course-specific knowledge to develop strategies to support patients in their last year of life and their families.*

Discussion

In this study, we used a realist evaluation approach to gain a deeper understanding of the particular contexts that may be specific to the experiences of young adults aged 16-40 years with cancer as they approached their end-of-life. We re-analysed our initial seven themes into seven CMO configurations that explained the specific needs of the end-of-life experiences of young adults with cancer. The implications of these specific needs are set out below and compared and contrasted with the existing literature.

Life course and not age matters

We found that within this group, end-of-life experiences and preferences cannot be neatly isolated into the two age ranges we studied (16-24 years and 25-40 years). A better way of approaching, understanding and supporting young adults may be to consider where they are in their life course, as there may be more in common, than different, between those with similar life course experiences, for example being in education, maintaining a career, having children or caring responsibilities.

The usefulness of taking such an approach is also found in the wider literature on end-of-life care for young adults and so reinforces this finding. Adolescence and young adulthood is a developmental stage when individuals shape their identities, gain autonomy, make career choices and develop intimate relationships. A cancer diagnosis at this stage is "off-time" during the normative life cycle: life is interrupted, developmental tasks and identity formation are challenged and few peers will share their cancer experience.(14) In common with Soanes and Gibson we found that participants across this age range reported a desire to maintain these aspects of their life, as well as their identity for example, as a student, a professional, or parent, in part to maintain a sense of normality and control.(15)

Giving young people the chance to have control and to feel normal

We found, perhaps unsurprisingly, the pivotal role of HCPs in supporting young adults with cancer. However, we were able to identify that an important 'block' to the support provided comes from the emotional discomfort felt by HCPs when discussing aspects of care specifically with young adults – such as discussions about prognosis. This is important as a cancer diagnosis creates great uncertainty and the knowledge that there will not be a cure creates a dissonance between the life that was expected and the reality of a life that will be significantly shorter than expected. For emerging adults and early independent adults, as disease progresses, dissonance is also present as their independence is compromised with an increasing and unanticipated dependence on others. This can affect their ability to attend school, college or work as well as taking part in family or social activities or fulfilling caring duties for others e.g. looking after young children. Adaptation is a mechanism through which there is a recognition of what can no longer be achieved due to disease progression.(16) An adapted normality can be achieved together with a sense of control, allowing for realistic goal setting.(17) Advance care planning could facilitate this adaptation. However, few participants in our study reported having had conversations about their options or the care they wanted to receive. Some HCPs avoided such conversations because of the emotional burden to themselves, not wishing to challenge either hope or a young person's possible denial about their situation. This might be an example of what Bell et al refer to ‘as social constraint’, i.e. words and actions that inhibit end-of-life discussions.(18) A further notable finding from the data indicates that all parties appear to wait for another to raise the topic of end-of-life. The ‘window of opportunity’ (17) often fails to appear, thus in some cases the topic is avoided. This has the potential to delay adaptation and limit the time available for professional support, which could help young adults plan and make as much as possible of remaining time.(19) For those with dependents, particularly young children, delaying adaptation could impact on their roles as parents, delaying the opportunity to prepare and create memories for themselves and their families.(17) When end-of-life was addressed, this tended to be when health had deteriorated, and that window of opportunity, albeit late, facilitated opportunities to discuss the future, end-of-life care and to make plans.

Families and carers matter even more

Data from family members came from two perspectives – both before and into bereavement. Many family members became informal caregivers. We found increased dependence on family members whether emotionally, physically, financially or for support with housing. The level of independence varied between the two age groups with those aged 16-24 more likely to be living in the parental home, still in education or receiving training and moving towards becoming independent from their family. Those aged 25-40 were more likely to have been independent adults for longer. In common with Knox et al, we also found that when thrust back into dependent relationships with parents, left behind by peers, whom they perceived to be moving forward with their own life goals, young adults could feel isolated.(20) The financial burden of cancer is widespread, but for those at the younger end of the age group who were still in education or living at home, the burden fell more heavily on their family. For those with greater independence and who relied on their income from employment, a cancer diagnosis compromised their ability to work and maintain their lifestyle. It is likely that older people with cancer, particularly those who have retired with an income to cover their regular expenses may not face such financial extremes. Mohammed et al. refer to caregivers ‘taking charge’, thrust into a role for which they often felt ill-prepared.(21) In our study, lack of understanding of the clinical situation due to confidentiality, a lack of practical or technical knowledge or skills and poor information from HCPs, themselves often reluctant to undertake end-of-life discussions, were some of the contextual influences contributing to feeling ill-prepared, abandoned or distressed.

The burdens for healthcare professionals

Professionals reported difficulty addressing the needs of both the person with cancer and their family as often they had different expectations. Professionals were aware that providing bereavement support to a family was difficult if they had not built a relationship with them in the limited time available. This is mirrored by our finding that continuity mattered much more to young adults. Managing complex family dynamics was challenging for HCPs and strategies to do this were often not addressed. Sometimes HCPs did not want to 'open a can of worms' by involving the family as they were aware that they would have to consider extra care needs, not viewed as part of their role. This was a strategy used by HCPs to manage their workload and families were not told that it was acceptable to ask for help and support. Professionals preferred to maintain and share optimism with the family, maintaining hope, all of which helped to reduce the emotional discomfort they would otherwise feel. So talking openly about the death of the person with cancer was rarely pursued. Beerbower et al. refer to 'a broken system of communication' that can lead to conflict, where there has been no disclosure of prognosis, or where disclosure has for some family members only been partial, or come much too late.(14, 22) Educating, enabling and supporting caregivers can thus be complex and challenging, reinforcing the need for early and developmentally appropriate communication.

Professionals often have less exposure to and experience of providing end-of-life care for young adults. They are likely to be similar in age to the person with cancer, their family or friends, enhancing the emotional difficulties of working with this population. Whilst in palliative care, end-of-life might be 'normal', caring for those aged 16-40 who are dying will not be normal nor will facing the loss of lives partially lived. The avoidance by HCPs of engaging in the challenging discussions and activities we have listed above is understandable. But Wiener et al. point out that HCPs need to reflect and be aware of the emotional effect that younger patients have upon them and whether the support they offer is relevant and enabling of this population to continue to live normally for as long as possible.(23) Clark et al. have suggested that providing a developmentally-appropriate approach to care that includes advance decision making is thus essential.(24)

To enable HCPs to meet the needs of the end-of-life care of young adults, formal support is needed. However, the formal support for HCPs in their professional roles varied in availability, access and was used differently. There was a distinction between doctors and nurses. Participants in our study suggested that the emotional burden received greater recognition in the nursing profession whereas for senior doctors there was little or no provision of support and an expectation that they would not show the emotional affect or their work. There were also issues about having the time to access support, associated costs and the lack of visibility and advocacy from senior HCPs for accessing support. In addition, support was not integrated into training or ongoing professional practice and for some senior HCPs it may have been regarded as compromising their role or authority.(25) Self-care in the palliative care workforce is known to be essential, yet rarely is education or training available.(26, 27) We would agree with Knox et al that palliative care services should consider prioritising resources to support self-care practice, to promote the health and well-being of HCPs.(18)

Strengths, limitations and future research directions

Although our study is unusual for the extensive data collected from young adults facing end-of-life and their triangulation with family and HCPs, recruitment of two groups of patients was unsatisfactory. Young adults with haematological malignancies were rarely invited to participate despite these being a commoner diagnosis in this population. This may be because those with haematological diagnoses continue to be offered and agree to receive 'curative' treatments.(28) When such curative options had been exhausted our participants were often 'actively dying' and too ill to participate in this study. Another under-represented group were those aged 16-24. Professionals suggested that whilst clinical teams identified young adults meeting the study eligibility criteria, the challenges of communicating that 'cure was not likely' may have increased the difficulty of introducing the study. Our original plan to undertake two interviews with participants failed: often patients were just too unwell for a second interview. Further research is needed to explore the needs of those often described as 'hard to reach', those with haematological cancers and those aged 16-24 years.

Conclusion

We identified challenges with the way current end-of-life care is delivered to young adults with cancer. Using this evidence, recommendations to improve care can now be developed.

References

1. Bleyer A, Ferrari A, Whelan J, Barr RD. Global assessment of cancer incidence and survival in adolescents and young adults. Pediatr Blood Cancer. 2017;64(9).
2. What are the top causes of death by age and gender? [Internet]. 2012 [cited 02 August 2016]. Available from: http://webarchive.nationalarchives.gov.uk/20160105221750/http://www.ons.gov.uk/ons/rel/vsob1/mortality-statistics--deaths-registered-in-england-and-wales--series-dr-/2012/sty-causes-of-death.html.
3. Globocan 2012: Estimated Cancer Incidence, Mortality and Prevalence Worldwide in 2012 [Internet]. 2012 [cited 02 August 2017]. Available from: http://globocan.iarc.fr/Pages/fact_sheets_population.aspx.
4. Ngwenya N, Kenten C, Jones L, Gibson F, Pearce S, Flatley M, et al. Experiences and Preferences for End-of-Life Care for Young Adults with Cancer and Their Informal Carers: A Narrative Synthesis. J Adolesc Young Adult Oncol. 2017;6(2):200-12.
5. Bleyer A, Barr R, Ries L, Whelan J, Ferrari A, editors. Cancer in Adolescents and Young Adults. 2nd ed. Switzerland: Springer; 2017.
6. Baker JN, Hinds PS, Spunt SL, Barfield RC, Allen C, Powell BC, et al. Integration of palliative care practices into the ongoing care of children with cancer: individualized care planning and coordination. Pediatr Clin North Am. 2008;55(1):223-50, xii.
7. Grinyer A, Barbarachild Z. Teenage and young adult palliative and end of life care service evaluation. London; 2011.
8. Kars MC, Grypdonck MH, de Bock LC, van Delden JJ. The parents' ability to attend to the "voice of their child" with incurable cancer during the palliative phase. Health Psychol. 2015;34(4):446-52.
9. Kars MC, Grypdonck MH, de Korte-Verhoef MC, Kamps WA, Meijer-van den Bergh EM, Verkerk MA, et al. Parental experience at the end-of-life in children with cancer: 'preservation' and 'letting go' in relation to loss. Support Care Cancer. 2011;19(1):27-35.
10. Pawson R, Tilley N. Realistic evaluation. London: Sage Publications Ltd; 1997. 235 ; 23 cm. p.
11. Wong G, Westhorp G, Manzano A, Greenhalgh J, Jagosh J, Greenhalgh T. RAMESES II reporting standards for realist evaluations. BMC Med. 2016;14(1):96.
12. Richards L, Qualitative S, Research. Using NVivo in qualitative research. London: Sage; 1999. ix, 218 pages : illustrations ; 25 cm. + 1 CD-ROM (4 3/4 in.) p.
13. Charmaz K. Grounded Theory: objectivist and constructivist methods In: Denzin NK, Lincoln YS, editors. Handbook of qualitative research. California: Sage Publications Ltd; 2000. p. 1065 ; 26 cm.
14. Beerbower E, Winters D, Kondrat D. Bio-psycho-social-spiritual needs of adolescents and young adults with life-threatening illnesses: Implications for social work practice. Soc Work Health Care. 2018;57(4):250-66.
15. Soanes L, Gibson F. Protecting an adult identity: A grounded theory of supportive care for young adults recently diagnosed with cancer. Int J Nurs Stud. 2018;81:40-8.
16. Livneh H. Psychosocial Adaptation to Cancer: The Role of Coping Strategies. Journal of Rehabilitation. 2000;66(6):40-50.
17. Nissim R, Rennie D, Fleming S, Hales S, Gagliese L, Rodin G. Goals set in the land of the living/dying: a longitudinal study of patients living with advanced cancer. Death Stud. 2012;36(4):360-90.
18. Bell CJ, Zimet GD, Hinds PS, Broome ME, McDaniel AM, Mays RM, et al. Refinement of a Conceptual Model for Adolescent Readiness to Engage in End-of-Life Discussions. Cancer Nurs. 2018;41(2):E21-E39.
19. Llewellyn H, Neerkin J, Thorne L, Wilson E, Jones L, Sampson EL, et al. Social and structural conditions for the avoidance of advance care planning in neuro-oncology: a qualitative study. BMJ Open. 2018;8(1):e019057.

20. Knox MK, Hales S, Nissim R, Jung J, Lo C, Zimmermann C, et al. Lost and stranded: the experience of younger adults with advanced cancer. Support Care Cancer. 2017;25(2):399-407.
21. Mohammed S, Swami N, Pope A, Rodin G, Hannon B, Nissim R, et al. "I didn't want to be in charge and yet I was": Bereaved caregivers' accounts of providing home care for family members with advanced cancer. Psychooncology. 2018;27(4):1229-36.
22. Bell CJ, Skiles J, Pradhan K, Champion VL. End-of-life experiences in adolescents dying with cancer. Support Care Cancer. 2010;18(7):827-35.
23. Wiener L, Weaver MS, Bell CJ, Sansom-Daly UM. Threading the cloak: palliative care education for care providers of adolescents and young adults with cancer. Clin Oncol Adolesc Young Adults. 2015;5:1-18.
24. Clark JK, Fasciano K. Young adult palliative care: challenges and opportunities. Am J Hosp Palliat Care. 2015;32(1):101-11.
25. Bradford NK, Greenslade R, Edwards RM, Orford R, Roach J, Henney R. Educational Needs of Health Professionals Caring for Adolescents and Young Adults with Cancer. J Adolesc Young Adult Oncol. 2018.
26. Mills J, Wand T, Fraser JA. Self-Care in Palliative Care Nursing and Medical Professionals: A Cross-Sectional Survey. J Palliat Med. 2017;20(6):625-30.
27. Mills J, Wand T, Fraser JA. Examining self-care, self-compassion and compassion for others: a cross-sectional survey of palliative care nurses and doctors. Int J Palliat Nurs. 2018;24(1):4-11.
28. Odejide OO, Cronin AM, Earle CC, Tulsky JA, Abel GA. Why are patients with blood cancers more likely to die without hospice? Cancer. 2017;123(17):3377-84.

Legends for figures

Figure 1. Phases of research process. Phases ii-iv are reported here.

Figure 2. Analysis process

Figure 1

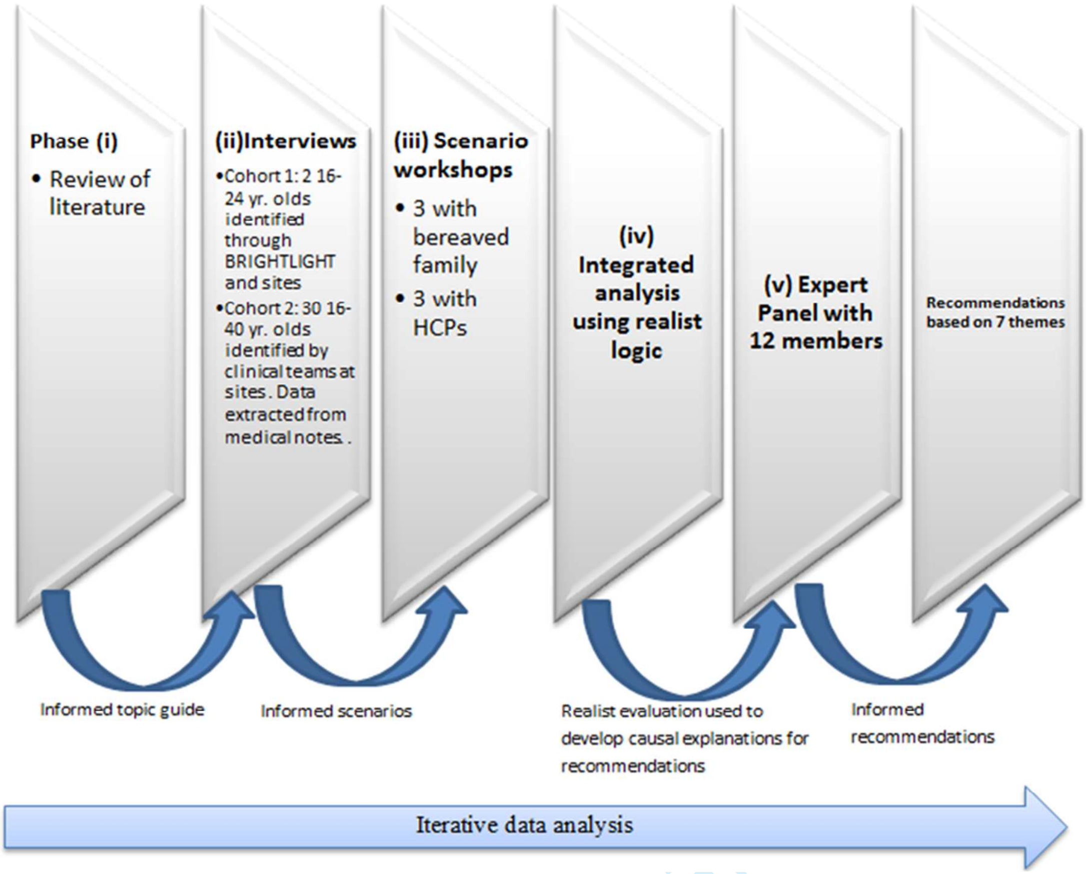
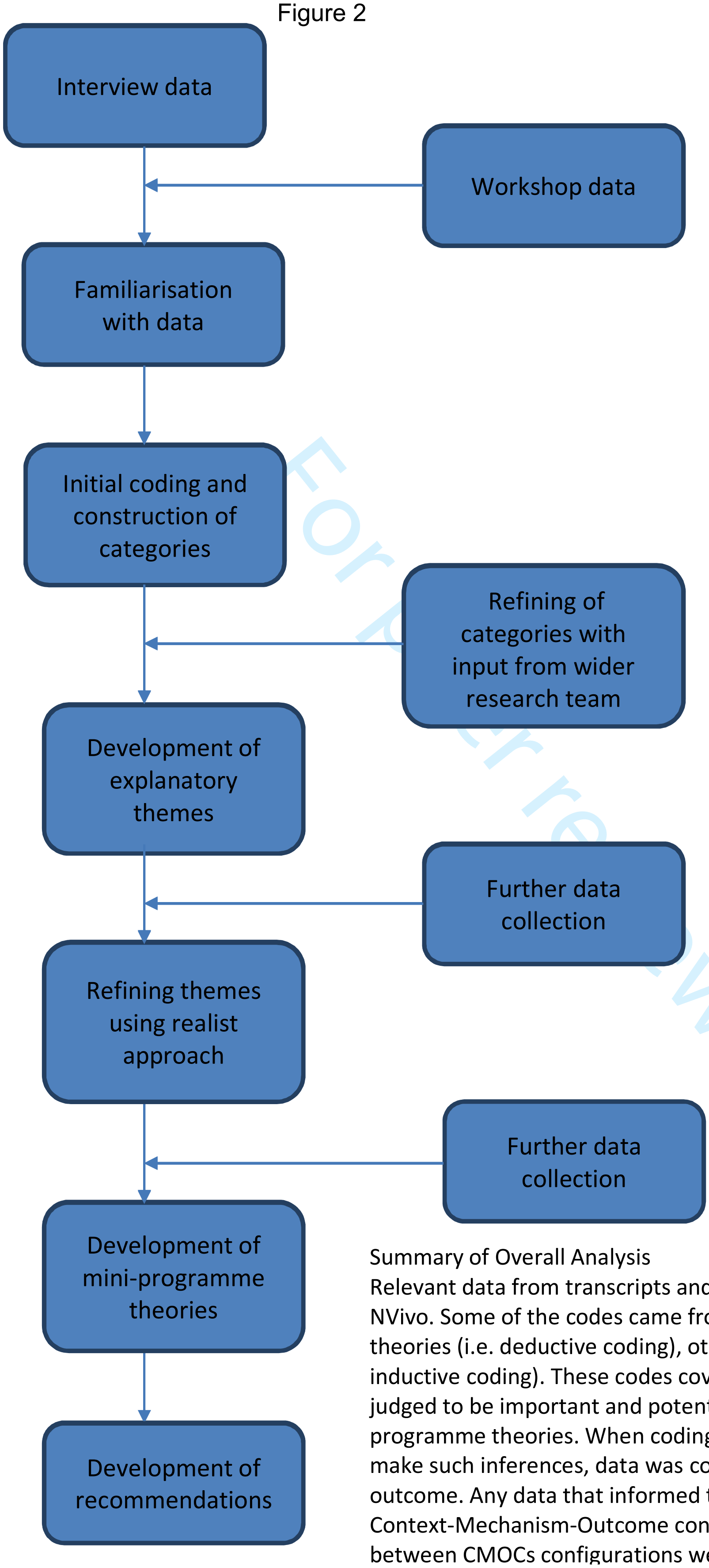

Phase (i)
• Review of literature
(ii)Interviews
•Cohort 1: 2 16-24 yr. olds identified through BRIGHTLIGHT and sites
•Cohort 2: 30 16-40 yr. olds identified by clinical teams at sites. Data extracted from medical notes. .
(iii) Scenario workshops
• 3 with bereaved family
• 3 with HCPs
(iv) Integrated analysis using realist logic
(v) Expert Panel with 12 members
Recommendations based on 7 themes
Informed topic guide
Informed scenarios
Realist evaluation used to develop causal explanations for recommendations
Informed recommendations
Iterative data analysis

Figure 2



Summary of Overall Analysis
Relevant data from transcripts and themes were coded into NVivo. Some of the codes came from the 'mini' programme theories (i.e. deductive coding), others from the data (i.e. inductive coding). These codes covered concepts that were judged to be important and potentially relevant to the 'mini' programme theories. When coding, where it was possible to make such inferences, data was coded as context, mechanism or outcome. Any data that informed the relationship of data within Context-Mechanism-Outcome configurations (CMOCs) or between CMOCs configurations were also coded.

Care towards the end of life in young adults with cancer - Figures

University College London Hospitals NHS
NHS Foundation Trust

Title of project: When cure is not likely: What do young adults with cancer and their families need and how can it best be delivered? A BRIGHTLIGHT companion study

PROTOCOL
Version 2.0 19th December 2013

Protocol Authorisation

PRINCIPAL INVESTIGATOR

Print name ………………………………………………………..

Signature ………………………………………………………..

Date ………………………………………………………..

Amendment History

Version	Date	Author	Application reference	Notes

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Lay Abstract

Background

This study will help increase our knowledge to understand better the needs of 16-40 year olds ("young adults") when cure from cancer becomes unlikely. The younger members of this group are on the borders of paediatric care and as cancer is relatively uncommon in this age group, patients are less often encountered in adult cancer and palliative care services. Overall cancer is relatively uncommon in people in early to mid-adulthood. Nevertheless, one quarter of deaths in 16-40 year olds are due to cancer. The impact of incurable cancer on patients and families is strongly influenced by age but for adults in early to mid-life, very little is known about their experiences as death approaches or how care is best delivered. Professionals consistently acknowledge many specific challenges of managing end of life care in this age group.

From this study we aim to understand;

a) The most important parts of care in the last year of life for people with cancer aged 16-40 years.
b) Whether differences exist between the experiences of people with cancer who are aged 16-24 and those aged 25-40 years.
c) How young adults and their families can be supported in the last year of life to achieve their preferences for care.
d) The challenges that exist for health and social care professionals providing care.

Methods

There are five distinct parts to our research.

I. **Analysing available information:** we will look at all available literature and information about end of life care in young adults including the information that is now collected routinely in the NHS about preferences and place of death.
II. **Interviews with patients, families and professionals**. We will work closely with health care professionals from four study sites (University College London Hospitals, Southampton University Hospitals, Leeds Teaching Hospitals, and St. Joseph's Hospice) to identify patients for interview about their care when cure is no longer likely. Fifteen 16-24 year olds taking part in the BRIGHTLIGHT study (a study already underway in the UK) and thirty 16-40 year olds from the four study sites will be invited to participate. We will ask them to propose a family member or carer and their key worker to be interviewed as well.
III. **Workshops with patients and professionals.** We will conduct nine workshops for interactive discussion and to increase our understanding of the range of perspectives, opinions and experience. Three workshops will be with family members and carers, three with professionals and three bringing together family members and carers and professionals.
IV. **Hold professional panels.** We will present our findings and recommendations to a panel of professional experts for further refinement.
V. **Make recommendations for practice.** We will have a good understanding of the core components and pathways of end of life care for young adults and make recommendations for practice and further evaluation to assist policy makers, commissioners and other stakeholders.

How the results of this study will be used

The findings will add evidence to inform national standards, pathways of care and core competencies for training staff. We will identify areas for change or further exploration. We will make public our findings widely to both lay and professional audiences.

1. Purpose of the study

This research will illuminate the core issues affecting end of life care[1] in young adults with cancer (aged 16-40 years), gathering evidence from the perspectives of the young people themselves, their families and the multidisciplinary team.

Using a national, multiple method realistic evaluation, we shall use an iterative approach guided by the MRC framework for evaluating complex interventions. [1,2] The aim is to define, describe and understand the core components for excellent practice in the delivery of end of life care for young people with cancer, to inform policy and practice and to set priorities for further evaluation studies.

In the United Kingdom (UK), health policy on specialist cancer services has bracketed young adults up to 24 years with teenagers (teenagers and young adults, TYA). [3] In the United States, Canada and Australia, strategies for improving cancer outcomes have most often focussed on 15-40 year olds ('adolescents and young adults', 'AYA'). [4] Notably, Douglas House, a unique hospice for young adults in Oxford serves an age range of 16-35 years. [5] This age group has attracted little attention in other European countries where there has been less consistency in the age range studied. We aim to explore comparisons between those aged 16-24 years and those aged 25-40 years to highlight key issues and differences that may be influenced by age.

In devising this research study, our underlying theories are:

1. That end of life care for young adults with cancer aged 16-40 years could be improved by increased understanding of (a) current care pathways for people with different types of cancer (b) the effects of age (above and below 25 years) (c) the need for accurate information (d) how active participation by young adults in decision making can impact on current and future care (e) the importance of respect for individual autonomy and family interactions.

2. That outcomes would be improved by an approach to end of life care that is (a) more aware of individual patient need and autonomy (b) supports professionals to recognise and respond to patient need (c) takes account of family interactions and relationships with patient and staff (d) enables patients to receive active and palliative treatments in a place of their choice to achieve their preferences for end of life care.

Thus, our objectives are to understand:

a) The core components in the pathways of care in the last year of life for people with cancer aged 16-40 years.
b) Any differences between the experiences of people with cancer from the age ranges 16-24 and 25-40 years.
c) How young adults and their families can be supported in the last year of life to achieve their preferences for care.
d) The challenges that exist for health and social care professionals providing care.

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[1] End of life care is defined as 'care that helps all those with advanced, progressive, incurable illness to live as well as possible until they die'. [6]

2. Background

One quarter of deaths in 16-40 year olds are related to cancer.[7] Overall survival rates have improved less than those of younger children and older adults.[8] Despite descriptions of the specific needs of teenagers and young adults for specialist cancer care, there is a dearth of empirical research, policy and expert practice related to their End of Life care. This is also true for young adults up to 40 years.

There are gaps in policy for this age group. Better care: Better Lives [9] makes no distinction between the needs of children and teenagers and young adults and deals exclusively with children's palliative services. Similarly the End of Life Care Strategy, Promoting High Quality Care For All Adults At The End Of Life makes no specific reference to young adults.[10]

Published literature about End of Life for teenagers and young adults has been confined to summaries of good practice or, where studies have been undertaken, data has been sourced from parents rather than young people themselves.[11-13] Others have undertaken retrospective analysis of medical notes [14] or produced comment and review papers.[15-18] Notably, just one study interviewed young people and explored their views on decision making as End of Life approached.[19]

Equally, work on the older young adult cancer population is limited. The few studies that have collected data from adults of all ages, either through interviews or questionnaires (EORTC QLQ-C30) have identified differences between ages at end of life including varying preferences for active treatment and greater symptom burden or lower quality of life.[20-22]

For those aged below 40 years, boundaries between curative and palliative treatment are often blurred; decisions may be influenced by cancer type, age and family circumstances. There is a lack of standardized models, approaches and communication aids for this age group; the management of symptoms and psychosocial concerns may be neglected if the challenges to communication are not overcome. Avoidance and delaying of discussions about End of Life decisions are common resulting in consequences for the quality of care.[15]

The challenges in delivering care to young people are as relevant to the delivery of end of life care as to intensive curative chemotherapy.[23-24] National policy in England and Wales directs that TYA have access to specialist services to meet specific challenges faced by young people in accessing services responsive to needs associated with the physical, educational, psychological and social developmental stages that are disrupted by the diagnosis and treatment of cancer.[4] This policy is being evaluated in the BRIGHTLIGHT study, (NIHR RP-PG-1209-10013; Appendix 3, study synopsis).

BRIGHTLIGHT will provide: a cohort of participants of all those aged 13-24 diagnosed with cancer in England in one year; the contexts and pathways of cancer care for young adults in England; sources of contributors to workshops and expert panels; prospectively collected patient outcome data and extensive analysis of the national cancer and end of life data sets. This takes into account information already available in the national cancer data repository (NCDR) and from the National End of Life Care intelligence network (Nend of lifeCIN) for those aged 16-40 years. BRIGHTLIGHT and this proposal are closely linked to the remit of the National Cancer Research Institute Teenage and Young Adult Clinical Studies Group.[25]

This study will provide the much-needed evidence to underpin quality of care and the enablers that will assist in addressing current barriers:

	Barriers to excellent end of life care	**Enablers of excellent end of life care**

When cure is not likely

We can influence	Evidence base: Poor understanding of (a) disease trajectory; (b) unmet health and social care needs; (c) variations between cancers; (d) variations between age, groups, gender, ethnicity Service factors: Support needs of staff , patients and families in decision making Attitudes and barriers to care of young people: structural; cultural; financial. Individual factors: personal and disease specific, education Costs of care	Data from BRIGHTLIGHT, NCDR and Nend of lifeIN Data from workshops and interviews Training and support health and social care professionals Understanding of the costs of components of end of life care components
What will influence our project	Recognition of last year of life and end stage disease Interaction between active and palliative treatments Family dynamics Attitudes of health care professionals National factors in healthcare environment; costs of care (perceived and actual), commissioning Regional variation in policy, service configuration and provision; cultural and economic factors Training and support needs of health and social care professionals	Integrated care pathways Strategic documents; DH and voluntary sector pressures; new service providers; public opinion; epidemiological trends Assessing transferability of intervention in regional sites Engagement of clinical champions; multi-disciplinary team approach to care Training and support for health and social care professionals

3. Study Design and Methods

Research will be undertaken in 5 discrete but inter-related phases.

- (i) Preparatory phase;
- (ii) Interviews with young adults, families and professionals;
- (iii) Scenario workshops with families and professionals;
- (iv) Analysis and interpretation of these data including synthesis with a literature review and quantitative data available through NCDR, Nend of lifeCIN and BRIGHTLIGHT then refinement through discussion with a panel of experts;
- (v) Development of recommendations for practice and further evaluation.

See Appendix 1 for diagram of these phases.

(i) Preparatory phase

This will be undertaken at UCLH and includes a detailed synthesis of the literature relevant to end of life for young adults with particular emphasis on identification and understanding of the mechanisms potentially causing the desired outcomes. Additionally, patterns of care described by National Cancer Data Repository and National End of Life Care Intelligence Network will be used to give further characterisation by definition of tumour types responsible for deaths, place of death and variations within our age range. This phase will inform the semi-structured realist interviews with young adults and scenario development.

(ii) Interviews

Several groups including patients, families and professionals will be interviewed as described below. Four sites will be involved in this part of the study.

We will conduct interviews with 45 young adults aged 16-40 years of age, purposively sampled to reflect a range of diagnoses (relevant groupings include leukaemia, lymphoma and solid tumours including brain, testis, sarcoma, carcinomas). Recruitment will be from the two sources outlined below (Appendix 2 illustrates recruitment, consent and data collection processes).

Sample 1 will be 15 teenagers and young adults aged 16-24 years participating in the BRIGHTLIGHT[2] cohort who will 'self-identify' through responses to trigger questions in the BRIGHTLIGHT survey indicating that a) no more treatment is possible, b) they have been offered/received care from the symptom control team or palliative care team or c) giving a response to the question asking what they had been told about their cancer suggesting that they are aware that cure is no longer likely. The BRIGHTLIGHT Senior Research Manager (SRM) or Cohort Manager (CM) will confirm with the young person's healthcare team that they are receiving end-of-life care (if this was not confirmed on the pre-survey check) and make sure that it is suitable to approach them to take part in the study. When this is assured they will call the young person, briefly describe the study and gain verbal consent to forward the information sheet. After approximately a week, the young person will be contacted via an appropriate means (e.g. 'phone, text) by the same person (SRM or CM) to see if they have received the information sheet and asked if they would like to take part in the study. For those who agree they will be asked for verbal consent to pass their contact details onto the RA who will then contact the young person.

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[2] See www.brightlightstudy.com for details of BRIGHTLIGHT and appendix 3 for a synopsis.

If a young person verbally consents to participate the RA will arrange a time and place to meet them for the interview. On the day before the interview the RA will call to confirm the young person would still like to participate; if they do then the RA will visit the young person as planned but before commencing the interview, s/he will confirm the young person understands what they are agreeing to and get written consent. For a detailed description of this recruitment process, refer to Appendix 4.

After a first interview, this group will be invited to participate in a second interview between 2 and 4 months later. At the time when the young person is recruited, they will be asked to nominate a key worker who will be in charge of providing support during the study. This key worker will be in contact with the researchers and will follow up with the young person after the interview to see if they need additional support.

Sample 2 will be interviews to include thirty patients aged 16-40 years. Recruitment will be facilitated by clinical staff at six study sites: University College London Hospitals, Southampton University Hospitals, Leeds Teaching Hospitals, St Gemma's Hospice, Wheatfields Hospice and St. Joseph's Hospice. These research sites were selected because they all have established palliative care teams and services, they actively participate in research, and the staff are skilled at recruiting patients in End of Life for research.

A poster will be displayed in appropriate spaces with contact details of the site PI and potential participants asked to talk to their clinician if they are interested. Once members of the clinical team have identified potential participants, they will make sure their details can be shared with the researchers. If the participant agrees, the researchers will contact them with more information about the study. Whenever possible, this will be done through a face to face conversation about the study, but if this is not possible, the information will be sent via post and questions will be answered over the telephone. The participants will then be given time to look at the information sheets and ask questions about the study. If the participant agrees to take part in the study, they will be asked to sign a consent form.

The young people in sample 2 will be asked to identify a family member, and a professional directly involved in their care such as the key worker who can be approached to be interviewed in addition (total 90 interviews). The young people will be asked to check with the family member to see if they agree that their details be passed to the researchers so that they can contact them with more information about the study. If the family member agrees, the researchers will contact them to talk about the study and provide them with the information sheet. The researchers will also approach the nominated healthcare professionals to provide them with more information about the study and to see if they are interested in taking part.

This second sample will allow targeting of patients identified by their key workers as being directly aware that cure is no longer likely and currently in the end of life phase. Ten will be aged 16-24, and 20 between 25-40 years. The sample size and distribution has been selected to add additional interviews to complement sample 1, to reflect the clinical heterogeneity present across this age range and in anticipation of saturation of themes during analysis (refer to Appendix 5 for the sampling matrix).

Using an investigator designed template (see Appendix 6), medical notes of interviewees will be reviewed to analyse written communication and documentation about discussions related to end of life in order to seek illustrations of best practice. Records of key discussions will be sought including communication between health professionals; record of advance care planning, do not resuscitate orders; communication about preferred place of death; insight of patient and family; information given and received; and evidence to show if limits of desired levels of information had been elicited. We will pilot the review of the notes at the beginning of the period of data collection and adjust the template to suit the content found in the medical notes.

All semi-structured realist interviews will be conducted by a researcher experienced in working with young adults and discussing sensitive issues. It is anticipated they will last for approximately one hour, will be digitally recorded, and transcribed verbatim prior to qualitative analysis of transcripts. Interviews will occur in the place of choice of participants, usually during clinic visits or in their own homes.

(iii) Workshops

Scenario development: end of life trajectories for young adults with cancer will be derived from an initial scenario development workshop attended by palliative care consultants, palliative care and cancer clinical nurse specialists, allied health professionals, oncologists and haematologists. Findings from the literature review, analysis of National Cancer Data Repository and National End of Life Care Intelligence Network data and emerging findings from the interview data will be presented. Scenarios will be co-constructed to precipitate discussion around key influencing variants such as diagnosis, different ages within the range being studied, symptom constellations and patterns of information disclosure and communication (for examples refer to Appendix 7).

Nine scenario workshops: A series of three workshops will be held in the study sites used in (ii) above using the scenarios to encourage interactive discussions and generate new thinking and potential solutions to problems raised. The use of different sites will support comparisons across contexts and address issues of generalisability and transferability of findings across different UK settings. Workshops at each site will be held with:

1. Families and carers of young adults with cancer; patient representatives and regional stakeholder organisation representatives (workshop maximum size 15)
2. All members of the multiprofessional team involved in delivery of treatment for cancer and end of life care (workshop maximum size 15)
3. Mixed participants representing groups (1) and (2) (workshop maximum size 20)

The family members and healthcare professionals will be recruited using two routes. First, from the participants who were interviewed as part of sample 2. The consent forms for the interviews explained in (ii) will include a section where the family members and healthcare professionals can indicate if they would like to be contacted to participate in the workshops. Second, additional family members and healthcare professionals will be recruited in the study sites by the researchers and members of the clinical team. Close contact with the clinical team will facilitate recruitment.

Consensus will not be sought, rather perspectives, opinions and experiences elicited allowing scenario ‘mapping’. This will be done in small groups using mapping aids to identify key ideas and hypotheses arising on how end of life care could be improved.

All workshops will be moderated by members of the research team who have experience of these methods as well as end of life/cancer care. Workshop teams will be developed to ensure consistency across this aspect of data collection.

(iv) Expert panel review

A panel of experts will be convened to include palliative care professionals, general practitioners, oncologists, haematologists, allied health professionals, policy makers, ethicists, charities and commissioners. Participants will not have been involved in previous workshops or interviews. Professionals will be identified by previous participants and members of the

research team. Professional organisations such as the NCRI Palliative and Supportive Care Clinical Studies Group will be consulted. These will be purposively selected based on their experience working with young people who are receiving end of life care, knowledge of policy issues and agree to participate.

A distillation of knowledge and understanding gained from realist interviews and workshops combined with quantitative data, will be presented to the expert panel describing ideal pathways with an explanatory account of key components of care. The panel will be asked to comment on areas of agreement and disagreement, provide alternative or additive explanations and to test and retest experientially the fit of mechanisms to potentially achievable outcomes as described in the pathways.

(v) Development of recommendations

The analysis and interpretation by an expert panel will lead to the writing and refining of an overarching explanatory account for end of life care for young adults with cancer. We shall present data to inform our objectives stated in section 1.

Emerging national standards, pathways of care and core competencies are anticipated and will be considered within the document. We shall circulate our draft to participants in the realist interviews and workshops, policy makers, commissioners and other stakeholders and take account of their comments in producing a final version. Identification of areas for potential intervention will be included and recommendations for future empirical evaluation and testing of the effect on outcomes will be suggested. Such empirical work would be the subject of the next phase in testing the feasibility and acceptability of the core components of delivering improved end of life care in a pilot observational study or exploratory randomised trial in the future. This would include consideration of the economic aspects of these approaches to care.

4. Data analysis

Methodological approach

We will use realistic evaluation, derived from critical realism. Its strengths are an emphasis on understanding the causal mechanisms which generate outcomes, consideration of context and a desire to improve practice and service delivery.[26] It supports a mixed method, iterative approach to capture multidimensional aspects of the evaluation of end of life care, which can be viewed as a complex, multi-component intervention. Realist research explores the links between context, mechanism and potential outcome. It increases our understanding of 'what works, for whom, in what circumstances, in what respects and why?' It seeks to penetrate beneath the observable inputs and outputs of an intervention. We shall initially untangle the influence of context in the care of young adults in age groups 16-24 and 25-40 years, with a range of cancer diagnoses, experiences and expectations of care. We shall use workshops to generate hypotheses on mechanisms by which care in the last year of life for people aged 16-40 years might be improved. Using qualitative data and quantitative data we shall assess which components of an intervention or approach to end of life care might lead to improvements in patient and family centred outcomes.

Analysis of interviews

Sample 1: Transcripts of the interviews will be analysed using a grounded theory approach. This provides a systematic and inductive approach for the collection of data, sampling and the building of theoretical frameworks.[27] Analysis will occur simultaneously with data collection. After reading and re-reading the transcripts, memoing and selective focused coding, constant comparison between codes will take place leading to development of categories. Software such as Atlas.Ti will support this. The second interviews facilitated by theoretical sampling will ensure completion of any conceptual gaps in the emerging theoretical framework with an understanding of individual experiences over time.

Sample 2: Grounded theory methods of analysis will also be used. The development of codes and constant comparison of codes in the development of categories will occur across the sources of data for each young adult. For example data will be triangulated using the interview data from the young person, their family member, nominated health professional and analysis of the medical notes. When a conceptual understanding has been developed for each case, analysis will occur across the sources of data as a whole.

Analysis of workshops

Detailed field notes will be taken, memos and post-it notes collected and workshop discussions will be audio-taped and transcribed verbatim as discussion around scenarios is as crucial as the 'mapping'.[28] Data will be entered into Atlas. ti. The research team will create a preliminary analysis from initial observations of the scenario maps developed and the transcripts of discussions from workshops (1) and (2) to generate a series of hypotheses of how end of life care can be improved. This initial analysis will be used to inform discussions in workshop (3).

After the data from the workshops is analysed, a short report will be distributed among the participants in order to share the findings as near to the time as data collection as possible.

Analysis for expert panel review

Analysis of data collected in an iterative process, a key aspect of realist methods, will be the focus. Preliminary thematic summaries of findings from the interviews and workshops will be combined with emerging quantitative data. Hypotheses on mechanisms of how end of life care could be improved will be carefully defined and prioritised. These will be refined further through discussion within the research team and with a panel of experts. This phase will allow a reconsideration of understanding of the interrelationships between the context and mechanisms generated by the experience of end of life care derived from all aspects of data collection, testing assumptions and exploring further remaining uncertainties.

5. Ethical considerations

We recognise that this is a sensitive area of research and we will be working with vulnerable participants. We anticipate the fact that the questions included in our interview could pose potential emotional and psychological burden for those involved. We are sensitive to this fact. The clinical members of our research team have significant experience in this area that we can draw upon. In addition we will make use of the now growing body of evidence of published studies on this topic.[29,30]

Therefore, we have taken the following measures in order to minimise the risks and burden for the research participants. The researchers to be appointed will have extensive experience of gaining consent and interviewing vulnerable participants. The researchers will ensure the protection and well-being of the participants throughout the entire duration of the study. The participants will be informed that they can ask questions or express their concerns about the study throughout its entire duration and can withdraw at any point. The researchers will also search for signs of discomfort or distress among the participants and will address them individually by talking to the participants and letting them know their options for withdrawal. During the discussion, the participants will be informed that they can refuse to answer questions. The information sheets contain the contact information of all of the members of the research team. The participants will be informed that they are free to contact the researchers with questions and concerns even after the study has ended.

Inbuilt support will be embedded in liaison with key members of the clinical team for recruitment, consent and support mechanisms for the participants if the interviews give rise to issues that need to be followed up (see Appendix 2). The researchers and the members of the clinical team will have ongoing communication about recruitment and data collection. The

members of the clinical team will play an instrumental role during the screening and selection process with the purpose of identifying potential participants and guaranteeing their protection. The key worker will be in charge of following up with the participant after the interview to see if any issues need to be discussed and provide appropriate guidance and assistance. This is important as the research may lead specific patients to face and discuss issues that had previously not been raised.

Considerable researcher burden is also possible. Having two researchers will ensure mutual support. There will also be extensive support from the research team, where expertise in methods and cancer care will be closely matched with researcher need and support. Monthly clinical supervision has been costed into our study.

6. User involvement

This aspect presents specific challenges and has been approached as follows: young adults and their families will be involved in data collection, feedback and analysis as an intrinsic aspect of our study; the NCRN Consumer Liaison Group has been approached to determine specific interest amongst members; the proposal will be read and commented on by family members of two young adults who died of cancer; input into patient information sheets and other patient information will be sought from the Young Peoples Reference Group associated with BRIGHTLIGHT (approximately 20 members - young people diagnosed with cancer between the ages of 14 and 25 (current age 18-29), some of whom have worked with us for over three years on various studies); additional input into patient and subject information sheets will be sought from an additional PPI group, the Cancer Partnership Research Group of the Surrey, Sussex and West Hampshire Clinical Research Network; presentations will be made from January 2013 to 'Kayleigh's workshop – terminally talkative' at the annual young people's conference, Find Your Sense of Tumour. This was first held in 2011 for those receiving end of life. Facilitated by two psychologists, it is now a regular event at the conference. The attendees from 2011 have also set up their own site on 'Facebook', moderated by psychologists, and which we can access if more immediate consultation on the study is needed. Through contact with the NCRN Consumer Liaison Group we have identified patients and patient representatives who will comment on the evolving study and contribute to workshops and panels.

We expect there will be very hard to reach participants consequent on limited awareness of End of Life, high levels of family and professional protection or exaggerated denial. Close relationships between researchers and key workers will be developed to improve chances of access. This will be enhanced by the clinical credibility and national profile of the research team.

7. Data sharing plan

BRIGHTLIGHT is approved by the National Information Governance Board reference ECC 8-05(d)/2011. Electronic data from the study will be stored on an NHS server supported by University College London Hospitals NHS Foundation Trust (UCLH). Paper documents will be stored in a locked filing cabinet in the cancer trials research facility at UCLH. Access will be limited to research associates and chief investigator. All digital recordings will be deleted once a written transcript has been produced. The interview and workshop transcripts will be anonymised and password protected. These will be archived, and available for further analysis after publication of the findings of BRIGHTLIGHT on End of Life.

8. Management of the study

The study is sponsored by UCL.
The core project team led by the CI work in close proximity and will meet regularly.
An advisory group will be established to regularly review and advise on study progress.

Timescales – total period 24 months

Month 1-4	Researchers start employment Detailed review of the literature and analysis of National Cancer Data Repository and National End of Life Care Intelligence Network Detailed protocol development
Month 5-17	Workshops and interviews
Month 18-21	Analysis
Month 21	Expert Panel
Month 22-24	Circulation of account to participants, policy makers, commissioners and other stakeholders. Completion of final account, dissemination through local meetings and writing of peer reviewed publications.

References

[1] Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M. Developing and evaluating complex interventions: the new Medical Research Council guidance. BMJ (Clinical research ed. 2008;337:a1655.

[2] MRC. Complex Interventions Guidance. 2008 [cited March 28th 2012]; Available from: (http://www.mrc.ac.uk/Utilities/Documentrecord/index.htm?d=MRC004871)

[3] National Institute of Health and Clinical Excellence. Improving Outcomes in Children and Young People with Cancer. 2005.

[4] Geiger AM, Castellino SM. Delineating the age ranges used to define adolescents and young adults. J Clin Oncol. 2011 Jun 1;29(16):e492-3.

[5] Helen and Douglas House. [cited 28th March 2012]; Available from: www.helenanddouglas.org.uk

[6] Department of Health. End of life care strategy. Promoting high quality care for all adults at the end of life. 2008.

[7] Office for National Statistics. Mortality Statisitics. 2010 [cited 28th March 2012]; Available from: http://www.ons.gov.uk/ons/publications/re-referencetables. html?edition=tcm%3A77-230730

[8] Barr RD. Adolescents, young adults, and cancer--the international challenge. Cancer. 2011 May 15;117(10 Suppl):2245-9.

[9] Department of Health. Better care: better lives. Improving outcomes and experiences for children, young people and their families living with life-limiting and life-threatening conditions 2008.

[10] Department of Health. End of life care strategy. Promoting high quality care for all adults at the end of life. 2008.

[11] Grinyer A, Thomas C. The importance of place of death in young adults with cancer. Mortality. 2004;9(2):114-31.

[12] Kars MC, Grypdonck MH, de Korte-Verhoef MC, Kamps WA, Meijer-van den Bergh EM, Verkerk MA, et al. Parental experience at the end-of-life in children with cancer: 'preservation' and 'letting go' in relation to loss. Support Care Cancer. 2009 Jan;19(1):27-35.

[13] Grinyer A. Palliative and End of Life Care for Children and Young People: Home, Hospice, Hospital. Oxford: John Wiley and Sons 2012.

[14] Cohen-Gogo S, Marioni G, Laurent S, Gaspar N, Semeraro M, Gabolde M, et al. End of life care in adolescents and young adults with cancer: experience of the adolescent unit of the Institut Gustave Roussy. Eur J Cancer. 2011 Dec;47(18):2735-41.

[15] Baker JN, Hinds PS, Spunt SL, Barfield RC, Allen C, Powell BC, et al. Integration of palliative care practices into the ongoing care of children with cancer: individualized care planning and coordination. Pediatric clinics of North America. 2008 Feb;55(1):223-50, xii.

[16] Christenson K, Lybrand SA, Hubbard CR, Hubble RA, Ahsens L, Black P. Including the perspective of the adolescent in palliative care preferences. J Pediatr Health Care. 2010 Sep-Oct;24(5):286-91.

[17] Pritchard S, Cuvelier G, Harlos M, Barr R. Palliative care in adolescents and young adults with cancer. Cancer. 2011 May 15;117(10 Suppl):2323-8.

[18] Wein S, Pery S, Zer A. Role of palliative care in adolescent and young adult oncology. J Clin Oncol. 2010 Nov 10;28(32):4819-24.

[19] Hinds PS, Oakes LL, Hicks J, Anghelescu DL. End-of-life care for children and adolescents. Seminars in oncology nursing. 2005 Feb;21(1):53-62.

[20] Lundh Hagelin C, Seiger A, Furst CJ. Quality of life in terminal care—with special reference to age, gender and marital status. Support Care Cancer. 2006 Apr;14(4):320-8.

[21] Parr JD, Zhang B, Nilsson ME, Wright A, Balboni T, Duthie E, et al. The influence of age on the likelihood of receiving end-of-life care consistent with patient treatment preferences. Journal of palliative medicine. 2010 Jun;13(6):719-26.

[22] Trevino KM, Maciejewski PK, Fasciano K, Greer JA, Partridge A, Kacel EL, et al. Coping and psychological distress in young adults with advanced cancer. Journal of Supportive

Oncology. 2011;http://www.supportiveoncology.net/single-view/aipcoping-and-psychological-distress-in-young-adults-with-advancedcancer/8ea0588008.html.
[23] Hollis R, Morgan S. The adolescent with cancer--at the edge of no-man's land. The lancet oncology. 2001 Jan;2(1):43-8.
[24] Whelan J. Where should teenagers with cancer be treated? Eur J Cancer. 2003 Dec;39(18):2573-8.
[25] National Cancer Research Network.
[26] Pawson R, Tilley N. Realistic Evaluation. London: Sage 1997.
[27] Charmaz K. Grounded Theory: objectivist and constructivist methods In: Denzin N, Lincoln Y, eds. The Handbook of Qualitative Research. 2nd ed. Thousand Oaks: Sage 2000:509-36.
[28] Oreszcyzn S, Carr S. Improving the link between policy research and practice: using a scenario workshop as a qualitative research toll in the case of genetically modified crops. Qualitative Research. 2008;8:473-97.
[29] Hinds PS, Burghen EA, Pritchard M. Conducting End-of-Life Studies in Pediatric Oncology. WJNR 29(4):448-465; 2007.
[30] Walshe CE, Caress AL, Chew-Graham C, Todd C. Case Studies: A Research Strategy Appropriate for Palliative Care? Palliative Medicine 18(8):677-684; 2004.

When cure is not likely

Appendices

Appendix 1: Phases of research
Appendix 2: Recruitment and data collection
Appendix 3: BRIGHTLIGHT study synopsis
Appendix 4: Sample 1 recruitment
Appendix 5: Sampling
Appendix 6: Review of medical notes
Appendix 7: Hypothetical scenarios

When Cure Is Not Likely - Methodology Protocol

Interviews and Workshops

[REC Ref: 13/LO/1098]

Rationale

This mini protocol outlines the approach to be implemented for the data collection phase (interviews, workshops and analysis). This will be informed by a realist approach which aims to understand the causal mechanisms which generate outcomes, consideration of context and a desire to improve practice and service delivery. Its iterative approach assists the capture of multidimensional aspects of the evaluation of End of Life Care and explores links between context, mechanism and outcome. It will increase our understanding of 'what works, for whom, in what circumstances, in what respects and why?'

WCINL: Aims and Objectives

Methods

Interviews

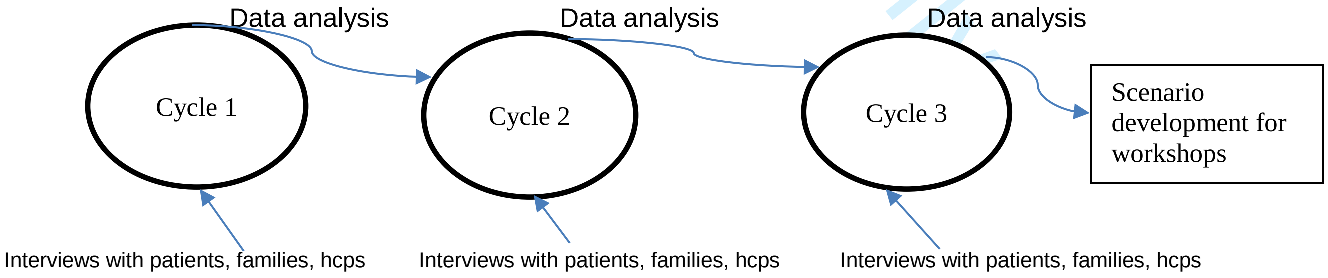
Realistic evaluation will be used as a guiding framework in exploring participants' experiences on the following outcomes;

Good patient centered care
Good family centered care
Responsive clinical care

Our understanding of what is implied by the term 'good care' will develop as our interviews take place and are analysed. For example, emotional spiritual physical; communication, information giving, management of symptoms; promotion of health family systems; social situation; quality of life; supporting finding meaning finding balance between acceptance and hope.

We plan a set of iterative cycles of interviews conducted with 3 groups in each cycle: patients, nominated family members and nominated health care professionals. We shall sample a maximum of 5 patients within each of the two age ranges (16-25 and 25-40) in each cycle, thus a maximum of 15 interviews per age group. Analyses of data will occur after each cycle and findings will be used to inform the conduct of the next cycle of interviews. Once data saturation is reached, the findings will be used to develop scenarios for use in the workshops in the next phase of our work.

Schema of iterative approach for interviews at Time 1:



The interviews will be explorative in their approach due to a significant lack of available evidence about the experiences of this population when cure is not likely and death approaches. This includes a lack of evidence on the availability and appropriateness of palliative approaches to care either alongside treatment or in the dying phase. Through the interviews we aim to explore and understand past and present experiences leading to how the future is viewed and conceptualized. We shall consider the internal, external and reflexive processes of the individual's experience. This will involve the different levels that impact on care: individual,

group (family and within healthcare professional teams), systemic and organizational (Ferlie and Shortell, 2001). We shall also explore the situational and contextual attributes for the individual.

The interviews will be iterative in their style using key questions and prompts to explore the perspectives and experiences of the participant. It is anticipated that each interview will last c. 1 hour. Interview schedules can be found at the end of this document, but in short;

> **Patients** will be asked to discuss their care, and perspectives of support for them, using the broad framework “What has happened, what is happening now, and what do you think will happen next?” Experiences of Internal processes; interpersonal and communication, relationship to others and the situation and context will be explored.
>
> **Family members** (nominated by patients) will be asked about the patient’s experiences as well as their experiences and perspectives of support for themselves as a family and/those in a caregiving role. This will include considering the effects on family dynamics, communication between family/ young person and professionals; how families can be supported to enhance their capacity to manage patient care and their perspectives of the past, present and future needs of the patient. Family centered outcomes in loss include experiences in bereavement [could use bereavement risk assessment tools for guidance (Agnew et al., 2010)].
>
> **HCPs** will be asked about their experiences of providing care for the specific patient who nominated them, their more general experiences of caring for patients in this age group when cure is no longer likely leading to discussions to inform our understanding of their internal processes of dealing/coping with younger patients who face death. Here the realist approach will allow both interviewer and interviewee to contribute to discussions of working in an area where sensitive issues are commonplace, thus facilitating opportunities for greater insights.

Patient Age Range: 16-25 years
Cohort 1: BRIGHTLIGHT
Sample size: 15 patients over 3 iterative cycles (maximum 5 patients per cycle)
Sampled from 4 cancer groups – minimum of 3 patients from each group

- leukaemia, lymphoma
- bone and soft tissue sarcoma
- brain and CNS tumours
- carcinomas including germ cell tumours

Interviews in this cohort will take place at time 1, and again after 2-4 months (time 2). The second interviews will explore the current situation and what has changed for the patient since the first interview using the same interview schedule but with a greater focus on what has changed. We anticipate that patients will have experienced both external and internal changes. We shall explore how they have experienced changes in their care, their relationships, and how they now conceptualise the future.

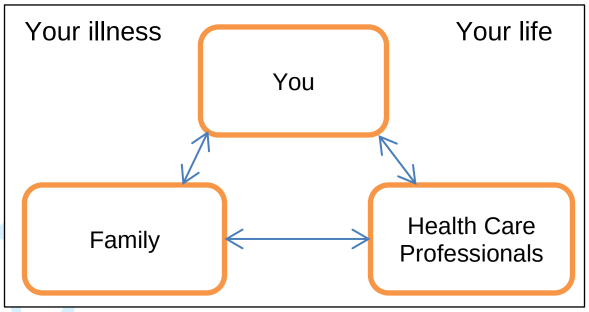
Patient age ranges: 10 16-24 yr olds; 20 25-40 yr olds
Cohort 2: Sites
Sample size: 30: interviews will occur at time 1 only.
Four cancer groups:

- Any carcinomas including breast, colorectal, melanoma, gynaecological and rare tumours – the most frequent diagnoses, so sample to recruit minimum of 21 patients
- Bone and soft tissue sarcoma – minimum 3
- Brain and CNS tumours – minimum 3
- Leukaemia, lymphoma – minimum 3

Conduct of semi-structured realist interviews:
A realist interview allows both interviewer and interviewee to contribute to discussions to facilitate deeper exploration of pertinent issues and allow the possibility of considering innovative issues and solutions.

Interviewers will work to a simple topic guide, allowing free discussion of issues in the past and present and what may occur in the future.

If a patient has difficultly answering questions or conveying their experiences a simple visual prompt may be used (example below) to provide a focus



Simple visual interview prompt

Analysis

All interviews will be audio-taped and transcribed verbatim. Two members of the research team will read in depth each transcript.

A modified grounded theory approach to analysis will be used (Charmaz, 2006). This will take an interpretative approach to identify themes and look for meanings and relationships within the interview data.

Each patient (cohort 2) will generate a set of 3 interviews which will be considered together as case studies which will then be compared and contrasted. In addition, the full set of each category of interviews (patient, family and HCP) will be analysed separately to extract common themes. We shall compare the datasets for the two age groups and look for commonalities and differences. Our findings will be used to inform the development of scenarios to be used in the workshops in the next phase of our research.

We shall also use our findings to develop further our programme theories. These theories have begun with the underlying hypotheses and enablers and barriers that we have identified at the start of our work from the literature and expert opinion of the research team. By defining our proposed outcomes as the provision of 'good' experiences of care for patients and families, and responsive care from HCPs, we shall use our data to develop a fuller understanding of what constitutes good and responsive care, and good experiences for patients and families. We shall use diagrams, flow charts, maps and memos to develop a full picture.

Outline of next stage of research:

Scenario workshops

Workshops will be held across our three recruitment sites across UK, Leeds, London and Southampton. Three workshops will be held at each site (total of 9) comprising of 1 with family members; 1 with healthcare professionals and 1 mixed family and HCP. The scenarios will be used to facilitate interactive discussion of issues arising for patients, families and HCPs in the experiences of care and illness for people in the age groups 16-25 and 25-40 years. The realist approach allows the opportunity for participant to disclose perspectives, opinions and experiences. The conduct of the workshops will enable the generation of hypotheses on mechanisms by which care in the last year of life for people aged 16-40 years might be delivered to enable 'good' care. We shall use our programme theories to underpin our thinking in the workshops and in the analysis of the data they generate. We shall use the emerging hypotheses to populate in more depth our existing programme theories. The teacher-learner approach by which researchers and participants both contribute to discussions will enable the researchers to use their theories and knowledge from the interviews to interact with workshop participants as well as confirm or falsify theories.

References

Agnew, A., Manktelow, R., Taylor, B. and Jones, L. (2009) Bereavement needs assessment in specialist palliative care: a review of the literature. *Palliative Medicine*, vol. 24:1, pp. 46-59.

Charmaz, K. (2006) *Constructing Grounded Theory: A Practical Guide Through Qualitative Analysis*. Sage, London.

Ferlie E.B, Shortell S.M. (2001) Improving the quality of health care in the United Kingdom and the United States: a framework for change. *The Millbank Quarterly*, vol. 79:2, pp. 281-315.

When Cure Is Not Likely – Interview Topic Guides

We need to collect some background information about each participant, so we can describe who has taken part in the study.
What year were you born?
How would you describe your gender?
Do you recall when you were first diagnosed (date/month/year?)
At the moment are you employed (on sick leave) or in full time education?
How would you describe your ethnicity?

During the interview, we would like to talk about a number of issues relating to your medical care, how this may have affected your social life, how well you think information has been given to you or how this could be improved and how decisions have been made about your care;

BOLD = questions Plain text = prompts

Patient	Past (up to WCINL) **Can you tell me what has happened up to this point?**	Present (WCINL) **What is currently happening?**	Future (Post Interview) **Do you think about things that might happen in the future?**
Medical Management	When did you find out something serious was wrong? [diagnosis] When did this happen, where, who was there, how did you feel What treatment did you have [if prompted by patient] How did you find out/ know things were not going so well [prognosis]	How do you think things are going with your illness /symptoms/ cancer at the moment? **Are you receiving any treatments now? Are you on medications, if so what are they for?** **What/who is helpful about the care you are receiving? What/who is unhelpful??** **How do you feel in yourself now? [feelings about medical aspects/situation]**	If yes, what do you think about / how do you plan?? Has anything been discussed with you about potential future treatment plans/ options [this needs to be dependent on what they say about the present]
Social Management	**How did you tell others (family/friends/colleagues) what was happening and how did they react** (illness, treatment, prognosis) **How did you find telling other people about your situation? If it was difficult, which parts were difficult??**	**Thinking about your day to day life – can you do the things that you want to do? If not, what is the impact on your life** **Does your illness have effects on you financially**?	**Is there anything in particular you want to do or achieve? (day to day; immediate, longer term)** **Do you have any worries/fears**

	On a very practical level, what were the biggest things that changed with the illness – e.g. where you were living / work / finance etc. **Have you used the internet to find out information about your illness** (What has been helpful/unhelpful? (illness, treatment) **Do you use social media like Facebook and Twitter? How has your illness affected that??**	**How does your current health affect you your relationships? (sexuality/family/friends/others/dependence/emotional)** **If you have a bad day, who/where do you turn to for support (what do they give you/how do they help? Have you found anything/ anyone particularly supportive)** **How do you feel in yourself now? [emotional wellbeing]** **Have you found a way to make some sense of what's happening at the moment? What has been helpful/unhelpful (spiritual)**	What are your fears (do you worry about) for the future? (parents/children/siblings, increase symptoms, being less able to do things you want to do, decreasing independence, dying process, life after death) **Do you have any hopes, dreams aspirations?**
Communication/information giving	**What was helpful / unhelpful about the conversations you have had with health professionals? How could it have been done better??** (e.g. timings / communication / information)	**Can you tell me about the support you are currently receiving from health care professionals (Cancer CNS, Pall Care, Consultant, GP, District Nurse, Social Worker)** **Can you tell me about the support you are receiving from your family (emotional, practical, financial, care)**	**What have your clinical team said about your future?** **What have you asked your clinical team about your future?** **Who do you think you would contact if you if you have concerns about the future? Why that person?**
Decision making	**Who has been involved in the decisions about your illness / care**?	**Are there any key decisions you are facing at the moment?**	**Are there any key things you think you will need to make decisions about in**

	What input have you had in decisions made about you illness/care?	**(further/stopping treatment; ACP; breaking news to others e.g. small children)** Prompt: If so, is anyone helping you with this? Is there anyone you would like to help you? Are you able to talk to that person / people… if not why not	**the coming weeks and months?** Who do you think might be able to help you with this? Are you able to talk to those people? – if not why not, **Are there questions you want to ask but don’t feel able to? What might help to make that easier??** Prompts: (Simple everyday things/activities; Preferred place of care; Preferred place of death; memory boxes, photos, Writing a will; Funeral planning)

Is there anything we haven’t mentioned that you would like to talk about today?

Family

We need to collect some background information about each participant, so we can describe who has taken part in the study.

How old are you?
How would you describe your gender?
Do you recall when you were first diagnosed (date/month/year?
At the moment are you employed or in full time education?
How would you describe your ethnicity?

During the interview, we would like to talk about a number of issues relating to x's medical care, how this may have affected their and your social lives and interactions, how well you think information has been given to x or to you and whether this could be improved and how decisions have been made about x's care.

Family	Past **What has happened up to this point**	Present **What is currently happening**	Future **What do you expect to happen in the future…**
Medical Management	When did you find out something serious was wrong with x [diagnosis] When did this happen, where, who was there, how did you feel **How involved have you felt in their treatment and care?** **How would you rate x's experiences of the care (diagnosis, treatment,;** helpful **/ unhelpful –in terms of care delivery / care pathway?** **Were you/other family members offered any support at this time by the clinical team (explain illness, treatment)** What was offered, did this differ by family members what was helpful/unhelpful	What are your views towards the current care that x is receiving How do you feel x is coping at the moment **How do you feel at the moment?** Do you feel supported Is there anyone who is supporting you? Do you have anywhere to go or anyone to talk to if you feel low? **How are the rest of your family coping (explore individual members)** **Is there anything more or different that could be done to help or support you or other family members?**	**Has anyone offered support to think about the future? If yes – was it helpful / unhelpful? If no – would you want some support ? who from? What should it look like?? What would you like?** What do you think might be available to help you and the patient in the future? (Pall Care, CNS, GP) What worries or hopes do you have about this?

	[if prompted by family member] How did you find out/ know things were not going so well [prognosis]		
Social Management	**What was the impact of x's illness on you/your family's day to day life (changes in relationships; priorities; carry on as 'normal')** In what ways has x's illness changed your family (closer, distant) **Who did you tell that x had cancer; how did they react; how did this make you feel?**	**What is the impact of x's illness on day to day life (financial, emotional)** How do you think this has changed your relationship with a) x b) other family members Have you found a way to make some sense of what's happening at the moment? How has this been? **Is there anything particular that sustains you when things are not going so well? (spiritual)**	**What are your fears/ hopes (do you worry about) for the future?** What support do you think you will need in the future (emotional, financial, care) What support do you think other family members may need
Communication/information giving	**What information were you given after x's diagnosis; who gave you this information; what did they say; what this information sufficient?** Do you think they were the best person to tell you this, if not, who would have been better How was information communicated between a) you and x; and b) between the different members of the family What was helpful/ challenging **Were there any things that you**	**What do you know about x's current situation** **What else would you like to know about their current situation; where/who would you go to find this out?** **Who do you turn to for support? Is it adequate, how could it be improved?? What should change??** Who do you share how you are feeling with (family, friends, counsellor); what do they provide	**Has x or their clinical team discussed with you about what might happen in the next weeks and months?** **Have you been offered any support for now or in the future? Is it adequate, how could it be improved? What should change?**

	couldn't talk about to some people; any people you could talk openly too?		
Decision making	**How involved were you in any decisions that x had to make about their treatment** Who decided on your level of involvement Would you have liked greater or less involvement?	**How involved are you in decisions that x has to think about or make (what are these decisions; what involvement have you had)**	**Have you thought about what will happen in the future?** Have you discussed the future with x; what have you talked about (Preferred place of care; Preferred place of death; memory boxes, photos, Writing a will; Funeral planning)

Is there anything we haven't mentioned that you would like to talk about today?

Heath Care Professional

We need to collect some information about each participant.
How old are you?
How would you describe your gender?
Could you tell me what your current job title is please?
How long have you been in this position?
How long have you worked in palliative care?
How long have you worked with the young adult population?
Have you completed any further training for working with young adults with cancer?

During the interview, we would like to discuss the past, present and future dimensions of care for x, and then talk about your wider experience of working with this patient population. With these topics in mind;

With reference to the patient:

- **Past:** How long have you known the patient; What has happened up to this point? (Diagnosis, treatment)
- **Present:** What does the patient/ family understand about what is happening; at what points has communication of significant issues around what is happening taken place, can you tell me more about these. What discussions about this patient have taken place across the MDT
- **Future:** What do you expect to happen in the future? (Prognosis, towards end of life); what has been put in place for the patient – ACP? What support do you think the family will need – has anything been put in place? What is difficult for the patient?
- What do you think would improve the pathway of care for this patient?? How should/could it be done differently?

Wider experiences of working with young adult population

- What are the most important aspects of care for this patient group? (holistic; pain/symptoms; family issues; socio-demographic; financial, Peers, life tasks, support, advocacy, listening, empathy compassion , being there)
- Beyond providing medical care, what other needs do this patient group often have and how do you identify and address these e.g. social, family, educational, financial needs do you think that your system is robust enough, or is there the potential for issues areas to be missed / forgotten?
- At what stage do you know 'when cure is not likely'; can you give me examples of breaking this news to a patient/their family - where it went well and an example of where it was more challenging. What can make these scenarios more difficult
- How do palliative treatments e.g. chemotherapy affect patients' desired end of life outcomes (Ref, USA evidence chemo in last months of life associated with CPR ventilation and dying in ICU, Wright et al, 2014)
- How do you negotiate patient/family relationships (e.g. family dynamics)
- What do you enjoy about working with this population?
- What do you find more difficult about working with this population

- How does it affect you when a relatively young cancer patient dies (emotional impact; coping mechanisms; specific patients e.g. those with young children)

- Have any personal or professional experiences affected your practice (e.g. using experiences to improve care; managing emotions; maintain a professional distance)

- Do you have clinical supervision/support available; do you use this or other sources of support? How else do you relax and gain perspective

- Any issues you are aware of generally for professionals, teams, units, places of care, caring for young adults with cancer who may die

- What could be put in place to improve end of life care for this specific patient group?

Are there any other aspects of care for this population that we have not discussed and you feel are important?

Reporting checklist for qualitative study.

Based on the SRQR guidelines.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the SRQR reporting guidelines, and cite them as:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. Standards for reporting qualitative research: a synthesis of recommendations. Acad Med. 2014;89(9):1245-1251.

		Reporting Item	Page Number
	#1	Concise description of the nature and topic of the study identifying the study as qualitative or indicating the approach (e.g. ethnography, grounded theory) or data collection methods (e.g. interview, focus group) is recommended	6-7
	#2	Summary of the key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results and conclusions	4
Problem formulation	#3	Description and signifcance of the problem / phenomenon studied: review of relevant theory and empirical work; problem statement	6-7
Purpose or research question	#4	Purpose of the study and specific objectives or questions	6-7
Qualitative approach and research paradigm	#5	Qualitative approach (e.g. ethnography, grounded theory, case study, phenomenolgy, narrative research) and guiding theory if	6-7

		appropriate; identifying the research paradigm (e.g. postpositivist, constructivist / interpretivist) is also recommended; rationale. The rationale should briefly discuss the justification for choosing that theory, approach, method or technique rather than other options available; the assumptions and limitations implicit in those choices and how those choices influence study conclusions and transferability. As appropriate the rationale for several items might be discussed together.	
Researcher characteristics and reflexivity	#6	Researchers' characteristics that may influence the research, including personal attributes, qualifications / experience, relationship with participants, assumptions and / or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results and / or transferability	n/a – semi-structured interviews and workshops
Context	#7	Setting / site and salient contextual factors; rationale	7
Sampling strategy	#8	How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g. sampling saturation); rationale	7-8
Ethical issues pertaining to human subjects	#9	Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	10
Data collection methods	#10	Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources / methods, and modification of procedures in response to evolving study findings; rationale	7-10
Data collection	#11	Description of instruments (e.g. interview guides,	7-10,

instruments and technologies		questionnaires) and devices (e.g. audio recorders) used for data collection; if / how the instruments(s) changed over the course of the study	supplementary upload
Units of study	#12	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	10
Data processing	#13	Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymisation / deidentification of excerpts	9
Data analysis	#14	Process by which inferences, themes, etc. were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale	9
Techniques to enhance trustworthiness	#15	Techniques to enhance trustworthiness and credibility of data analysis (e.g. member checking, audit trail, triangulation); rationale	9-10
Syntheses and interpretation	#16	Main findings (e.g. interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	12-16
Links to empirical data	#17	Evidence (e.g. quotes, field notes, text excerpts, photographs) to substantiate analytic findings	12-16
Intergration with prior work, implications, transferability and contribution(s) to the field	#18	Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application / generalizability; identification of unique contributions(s) to scholarship in a discipline or field	21-25
Limitations	#19	Trustworthiness and limitations of findings	25

Conflicts of interest	#20	Potential sources of influence of perceived influence on study conduct and conclusions; how these were managed	25
Funding	#21	Sources of funding and other support; role of funders in data collection, interpretation and reporting	Cover sheet and upload

This checklist can be completed online using https://www.goodreports.org/, a tool made by the EQUATOR Network in collaboration with Penelope.ai

Understanding care when cure is not likely for young adults who face cancer: a realist analysis of data from patients, families and healthcare professionals.

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Key words: Qualitative, Adolescents, young adults, cancer, end of life care

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Author Contributions

CK, NN, FG, MF, LJ, SP, GW, KMB, SH, RH, AH, LCS, RMT, AT, JW were all involved in study design, review of results and review of this manuscript. Interviews and analysis were conducted by CK and NN: aided by data analysis meetings with LJ, SP, FG, and GW. Workshops were attended by CK, NN, MF, LCS, RH, FG, JW: KB and ABH attended 1 workshop and led another. Participant recruitment was assisted by MF, KB, SH, AH, LCS, JW.

Data sharing statement

No data from interviews with patients, families and health care professionals is available in the public domain given the potential for identification of sensitive information.

Abstract

Objectives: To understand the experiences of young adults with cancer for whom cure is not likely, in particular what may be specific for people aged 16-40 years and how this might affect care.

Design: We used data from multiple sources (semi-structured interviews with people with cancer, nominated family members and healthcare professionals, and workshops) informed by a preliminary programme theory: realist analysis of data within these themes enabled revision of our theory. A realist logic of analysis explored contexts and mechanisms affecting outcomes of care.

Setting: Three cancer centres and associated palliative care services across England.

Participants: We aimed for a purposive sample of 45 people with cancer from two groups: those aged 16-24 years for whom there may be specialist cancer centres and those 16-40 years cared for through general adult services; each could nominate for interview one family member and one healthcare professional. We interviewed three people aged 16-24 years and 30 people 25-40 years diagnosed with cancer (carcinomas; blood cancers; sarcoma; central nervous system tumours) with an estimated prognosis of less than 12 months along with nominated family carers and healthcare professionals. Nineteen bereaved family members and 47 healthcare professionals participated in workshops.

Results: Data were available from 69 interviews (33 people with cancer, 14 family carers, 22 healthcare professionals) and six workshops. Qualitative analysis revealed seven key themes: loss of control; maintenance of normal life; continuity of care; support for professionals; support for families; importance of language chosen by professionals; financial concerns.

Conclusions: Current end-of-life care for young adults with cancer and their families does not fully meet needs and expectations. We identified challenges that are specific to those aged 16-40 years. The burden that care delivery imposes on healthcare professionals must be recognised. These findings can inform recommendations for measures to be incorporated into services.

Strengths and limitations of this study

1. There is a lack of empirical research, policy and expert practice to inform delivery of optimal care for young adults, and support of their families, when cure of their cancer is not likely.
2. Data were therefore collected by interviewing young adults under 40 years of age with incurable cancer, their nominated family carers and healthcare professionals.
3. Further primary data were obtained in workshops with bereaved family members and professionals involved in end-of-life care for young adults.
4. People with blood cancers and those aged between 16-24 years were difficult to recruit and may have unrecognised specific needs.
5. Analysis of this unique data set has highlighted specific challenges for young adults, their families and healthcare professionals in the delivery of end-of-life care.

Background

Cancer in young adults under 40 years is notable because it comprises a wide range of malignancies, has specific challenges to improving both length and quality of life but is relatively uncommon.(1) One quarter of all deaths in the United Kingdom in people aged 16-40 years are from cancer.(2) In Europe there are over 27,000 deaths per year in this age group.(3) Despite increasing empirical evidence of the specific needs of young adults in specialist cancer care, there is little evidence about their experiences at the end-of-life.(4, 5) Studies of adults with cancer usually cover a wide age range with most participants aged over 40 years. The existing literature tends to summarise good practice and, where studies have been undertaken, little evidence comes directly from people with cancer. (6-9) Given the identified gap in current literature, this research aims to contribute to Ngwenya et al.'s conclusion that “Future research should focus on age-specific evidence about the end-of-life experiences and preferences for young adults with cancer and their informal carers”. (4)

Boundaries between curative and palliative cancer treatments are often blurred as decisions may be influenced by cancer type, age and family circumstances as well as the experience and skills of healthcare professionals (HCPs). Avoidance and delaying of discussions about end-of-life decisions are common, often affecting the quality of care.(10) Professionals consistently acknowledge the challenges of managing end-of-life care for people in this age group, which may have commonalities with and, importantly, differences from those people with cancer at other ages as death approaches. (8, 9, 11)

To develop our knowledge of end-of-life care in adolescents and young adults aged 16-40 years (referred to in this paper as ‘young adults’) with cancer, we sought to collect data directly from young adults who were facing a poor prognosis, their families and HCPs involved in their care. To gain a deep understanding of the contexts that may be specific to this age group, we chose to explore our data using a realist evaluation approach.(12) A realist evaluation approach focuses on explanations, taking account of contexts and mechanisms that may affect outcomes. It addresses questions about what works for whom, in what circumstances and in what respects, and how?

Consistent with the realist evaluation approach , we began our research with a preliminary programme theory. A programme theory is a description, in words or diagrams, of what is supposed to be done in a policy or programme (theory of action) and how and why that is expected to work (theory of change). (13) Details about how to develop programme theories is beyond the scope of this paper but methodological guidance is available.(14) Our preliminary programme theory was informed by expert opinion within our research team which was led by clinical academic specialists in the care of young adults with cancer. Our thinking was also informed by a narrative review of the existing literature, Phase i of our study, previously reported.(4)

Our preliminary programme theory was:
‘That there are specific differences in experiences of and preferences for care towards the end-of-life for those with cancer aged 16-24 and 25-40 years compared to those who are older. Life-threatening illness in the young is untimely, it disrupts expected biographies, and maintaining a sense of control and normality in everyday life may be important. The role of close family members is complex and integral to the experiences of the person with cancer.’

We used this theory to develop topics for use in semi-structured interviews with young adults with cancer, family members and HCPs, and to underpin scenarios used in workshop discussions with HCPs and bereaved family members. That is, our preliminary programme theory sets out our initial hypotheses of the differences we thought were likely to set apart the end-of-life care experiences and preferences for younger people. Our interviews were thus developed by the project team in such a way as to be able to gather data that would enable us to confirm, refute or refine aspects of our programme theory. For example, because we hypothesised that a sense of control might influence end-of-life care experiences, we deliberately developed interview questions that asked about this issue. An important point about our initial programme theory is that it was refined as the evaluation progressed based on data gathered. As such, our expectation was that our preliminary programme theory would need to be refined to have adequate explanatory value.

In this paper, we describe data arising from these interviews and workshops. We used our data analysis to further explore and develop realist causal explanations that may explain parts of our preliminary programme theory. As is expected in realist evaluations, as the evaluation progressed, we developed a revised programme theory that can be used to underpin recommendations for policy and practice and inform future research.

Methods

A multi-method realist study was undertaken (Figure 1). A realist evaluation approach was used as we wanted to explain and understand contextual influences on the experiences of and preferences for care towards the end-of-life for those with cancer aged 16-24 and 25-40. Here we report on Phases ii-iv, using RAMESES standards for reporting realist evaluations.(15) Phase v will be reported separately.

Recruitment and participants

We aimed to recruit a purposive sample of 45 people aged 16-40 with cancer, in two cohorts with an expected prognosis of less than one year, across four cancer groups: carcinomas; leukaemia and lymphoma; bone and soft tissue sarcoma; and central nervous system (CNS) tumours, which account for more than three-quarters of cancers occurring in this age group. In cohort 1 we planned to recruit 15 participants aged 16-24 years, to be interviewed at two time points; recruitment began via a national cohort study investigating whether specialist cancer services add value (www.brightlightstudy.com) and was later extended, due to poor recruitment, to include five principal treatment centres and a hospice for young adults. Cohort 2 was recruited from three specialist cancer services and three hospices in England and consisted of a purposive sample of 30 participants between the ages of 16-40. All cohort 2 participants were invited to nominate a family member and HCP involved in their care for interview. The first-hand clinical experience of many in the project team aided the development of the study. Knowing that this is an under-researched population within the context of the study and drawing on professional experience to guide data collection, analysis and interpretation was essential. Further details are available in the protocols (supplementary files 1 and 2).

Data collection

Semi-structured interviews

All participants took part in a semi-structured interview at a single time point using a topic guide. Cohort 1 participants were invited to take part in a later second interview.(12) The topic guide was developed from a review of the limited existing literature for the 16-40 age range (4) and the clinical and academic expertise within the project team who work directly with this population. We sought patient and public involvement (PPI) input to refine the topic guide coverage and phrasing of the questions, which explored medical, social, communication and decision-making experiences for people with cancer and their families. We asked HCPs to reflect on the care of the person with cancer and their practice with those approaching the end-of-life.

Workshops

We held workshops in London, Southampton and Leeds. The workshops involved the participants sitting as one group. One clinical member of the team acted as the facilitator for the HCP workshops and two clinical members of the team were co-facilitators for the bereaved relative workshops. The co-facilitation meant that if someone from the group needed to leave or have a break from the discussion they could be supported by one of the co-facilitators whilst the workshop was able to continue. At the start of the workshop the facilitator introduced the study, outlined the workshop and informed consent obtained. The HCP workshops focused around the scenarios and the perspectives of different professional roles. The bereaved relative groups were guided by one of the facilitators with the participants sharing narratives around their experiences with other participants either supporting the narrative or outlining how their experience differed.

1. Healthcare professionals

Three workshops involved HCPs working in both hospital and community settings who were recruited by the participating sites. Two scenarios were developed from initial interview analysis and reported experiences (Table 1). We sought to present contrasting fictional patients differing by age, gender and social situations which had raised a number of common issues arising from the interview data that the workshop participants were asked to discuss.

2. Bereaved relatives

We held three workshops with bereaved relatives who were invited to take part by bereavement services in participating hospices. The use of scenarios for this group were felt to be too abstract; and so these workshops focused on the relatives' individual experiences. The workshops involved open discussions and sought to collect information that had not emerged previously in the interviews, particularly concerning the last days of life.

Table 1. Scenarios used in health care professionals' workshop

Scenario 1: 16-40 year old patient with haematological malignancy	**Scenario 2: 25-40 year old patient with oncological malignancy**
Mannu, 19, diagnosed with Hodgkin's disease in December 2013. Between December and June treated with curative intent. Relapsed June 2014 – no sibling bone marrow donor available – deteriorated before one could be found. <u>Social</u> Science student – sporty. University not local. Friends all at University. School friends all over country also at University. Keeps in touch with friends via Facebook. <u>Home</u> Returned to live with Mum, Dad and sister aged 12. Grandparents supportive – all aware of diagnosis and prognosis. Sikh faith. Supportive in background. Home is a three bed semi with a bathroom upstairs and downstairs toilet. <u>November 2014</u> Inpatient. Deteriorating – wants to be at	Helen, 38, diagnosed with colon cancer in May 2014. Helen lives with her partner and their 18 month old baby. Soon after diagnoses she had surgery for a stoma fitting and was diagnosed with liver metastases a few weeks after. <u>Social</u> She has support from her parents, brother and her partner's parents. She is currently on sick leave and misses friends from the office. They have reduced income due to her being on maternity leave before her diagnosis, although she has critical Illness Policy which will pay off their mortgage and so this is reassuring for her. <u>Home</u> She lives an hour's drive from her parents in a duplex house with stairs. She is getting more symptomatic and experiencing fatigue. Partner is concerned about coping with a

home. Unable to do stairs therefore need to make adaptations. • Symptoms – shortness of breath, cough and fatigue. • Care – family keen to do. • Discharge home with Community Palliative Care Team input. • Contact with charities - Willow Foundation, CLIC Sargent <u>December 2014</u> Increased fatigue. Treated with radiotherapy to chest. Cough and fatigue. <u>January 2015</u> Further deterioration. Bed bound. Home oxygen. Anticipatory medications.	young child and partner as she deteriorates. <u>September 2014</u> Helen has lost weight and is aware that she is getting weaker and has difficulty picking up/carrying her child. She is currently on a 24/7 syringe driver and the District Nurse visits daily. Referral to hospice palliative care has been made but she has not yet been in contact. Helen is referred for a clinical trial as still relatively well and no conventional treatment options. <u>December 2014</u> Chemotherapy stopped as disease not responding - parents devastated. Parents not able to access psychological support as they live 'out of the area' Advanced care planning with clinical nurse specialist causes tension as parents do not wish Helen to be 'not for resuscitation' <u>February 2015</u> House requires adaptations due to her physical condition. Increasingly housebound due to steps and steep hill Partner feels he can no longer cope as Helen's condition deteriorates further.

All patient, family and HCP participants were provided with a Participant Information Sheet which outlined the study, their expected involvement and the right to withdraw at any point. Written informed consent was obtained from all those who participated in the study. Interviews and workshops were audio-recorded, transcribed verbatim and anonymised before analysis. Field notes were recorded during the workshops.

Data Analysis

Data were entered into a qualitative analysis software programme, NVivo 10 to facilitate analysis. (16) A realist evaluation approach enabled us to identify and understand (a) the outcomes for young people receiving care; (b) when these outcomes were likely to occur (the contexts); and (c) why (the mechanism).(12) Our analysis was multi-staged (figure 2):

- *Stage One - identification of emergent themes.* Charmaz's approach was used.(17) Initial codes (summary of what participants were describing) were open and inductive from the data using verbatim quotes or researcher-generated codes to inform a conceptual framework. We then developed categories by grouping similar codes. The categories were discussed and further refined into themes.
- *Stage Two - realist logic of analysis.* This stage was undertaken as we wanted to develop findings that had a clear warrant for transferability. In other words, by

reanalysing our themes, using a realist logic of analysis, we would be able to identify the commonly occurring mechanisms within this population group that caused the outcome patterns we had found. The way we operationalised a realist logic may be found in supplementary file 3.

Reinterpretation of the themes was undertaken by CK and NN aided by data analysis meetings with LJ, SP, FG, and GW. To assist the reinterpretation process, we developed 'mini' programme theories that explained the care pathways and experiences of the patients, family members and HCPs. For each of these mini programme theories we re-analysed the data that we drew on to develop each theme to build context-mechanisms-outcome (CMO) configurations - i.e. develop realist causal explanations of outcomes that occurred within different contexts (e.g. social rules and cultural systems). Workshop data were analysed in the same two-step manner and used to confirm, refute or refine the CMO configurations within the 'mini' programme theories.

Ethical review

The study was approved by Central London Research Ethics Committee (Reference: 13/LO/1098) and informed consent was sought from all participants at the time of participation.

Patient and Public involvement

We responded to a funding call from Marie Curie, a UK charitable organisation which provides care and support to people with terminal illnesses and their families, specifically seeking research proposals focussed on the needs of young adults. We sought the views of people with cancer on study design and written information including patient information sheets through the Cancer Partnership Research Group of the Surrey, West Sussex and Hampshire Cancer Network and the National Cancer Research Network Consumer Group. An independent steering committee, which included a bereaved parent of a young adult, provided advice and oversight on study conduct. We plan to work with Marie Curie on patient-focussed dissemination of our findings.

Results

Table 2 summarises the participants by cohort. A total of 69 interviews were conducted (33 people with cancer, 14 family members, 22 HCPs); 19 bereaved family members and 47 HCPs took part across six workshops.

Table 2 Participant Details

Cohort 1 N=30		Cohort 1 N=30	Cohort 2 N=3
Gender	Male	11	3
	Female	19	0
Age	Median (range) years	32 (16-39)	
Ethnicity	White British	19	3
	Any other White background	4	
	Asian/Asian British/Black/African/Caribbean/Black British	7	
Cancer type	Carcinoma	18	1
	Sarcoma	6	2
	Blood cancer	2	
	Other (incl. melanoma/CNS)	4	
Education/Working	Working Part Time	2	
	Working Full Time	2	
	Sick Leave	9	2
	Sick leave from education	2	1
	Not Working/Early retirement	14	
Nominated, interviewed family or other	Husband/Wife/Partner	5	
	Parent/sibling	8	
Nominated, interviewed healthcare professionals	Clinical nurse specialist	13	
	General Practitioner	2	
	Hospital doctor	4	
	Allied Health Professional	3	
	Patient did not nominate	5	
	Healthcare Professional declined participation	3	

The results are presented in three sections:

1. Our thematic analysis of qualitative participant data.
2. Realistic logic of analysis reporting context, mechanisms and outcome (CMO) configurations developed from re-analyses of the themes.
3. The connections and links between contexts, mechanisms and outcomes as leading to the revision of our programme theory.

Section 1 Thematic analysis

Seven key themes emerged each of which is accompanied by one or more illustrative verbatim section of texts from our data.

Loss of Control

As illness progressed and young adults with cancer became more debilitated, they often felt a loss of control over how they lived their lives. This was a shift from independence to a growing dependence on others for physical, emotional, practical or financial support provided by family, friends, HCPs or the wider state. The future became unpredictable and planning was difficult. Maintaining a sense of control and continuing to take part in activities, albeit compromised, was important:

My independence. For me, being able to do things on my own is definitely something that I miss, without - being carefree, I can't be carefree, I can't just go out and have, get drunk with friends any more. I can't go out for a long night and dress up in heels and get bashed about, because I have a port in, I've got cancer, you know, I have to go and sit down at a bar, have a non-alcoholic cocktail. It doesn't mean I can't socialise and have a good time with them, I still do. But I'm uncomfortable when I dress up now, whereas before I had the figure and went to the gym and felt more comfortable in myself. (Cohort 2 – Patient 20)

Maintenance of Normal Life

Participants all desired to continue, as far as possible, living a 'normal life' e.g. working, taking part in activities, looking after their children. Normality provided reassurance and a sense of control but it could also be a defensive response and a shield of denial about the realities of dying from cancer. As the disease progressed the sense of 'what was normal' needed to be reframed and adjusted:

I'm at probably the worst stage I've ever been with this illness, obviously because it's more advanced. Yet people are just saying, "You're looking great." And when I look in the mirror, I don't feel like I've got cancer. I don't feel like – obviously I do because I know that I do, but I don't feel any different to how I used to feel. Obviously yes you've got a few aches and pains and stuff, but you think like, when you hear someone's dying of cancer, you think that person will feel like they are. But like I know that I am, but I don't feel like I am, because it's quite a disconnect of like how – you know, like when you're feeling alright and you're going round doing stuff, and you're just doing stuff like everyone else, you just kind of forget. You go to work and you just have the same sort of, do the same things you were doing before you had cancer. You just forget, I forget sometimes (Cohort 2 – Patient 19)

Continuity of Care

Young adults valued being known by the HCPs involved in their care and preferred a joined-up care pathway between them, the HCPs and other health services. This relied on maintaining continuity of communication and information between HCPs, services and themselves with a shared knowledge of the care plan. They generally preferred to be seen by the same HCPs as they felt they could build rapport and feel known as a person. When they moved between services e.g. from oncology to palliative care or from hospital to hospice, they wanted this to be a joined-up seamless shift:

So we went into this initial meeting and [1st tumour CNS], who is the CNS, was there. And Dr [Consultant] was the one that kept us waiting. And it was said at that point, "[1st tumour CNS] will be your CNS, presumably key worker, throughout this process, she will be at every one of your appointments when you come to clinic." And I was like, great, and he gave me her number and a pack and, you know, I felt quite supported by that. … I understand not being able to the same nurse every time, that's not possible, but like if you had a team that were allocated a certain number of patients – because they just, they don't know you. And I've noticed that across the course of having another lot, you know, and I've really – I've kind of got to know a lot of them because I've been there, you know, over the course of a year. But, you know, it is at the beginning, it's someone different every week. And they don't know anything about you. And I went in expecting them to have read my notes, know what kind of cancer it was, know, you know, some of my background, and totally naively – they – and I think it's unfair to them, they are there just to administer medication (Cohort 2 – Patient 29)

Professionals Need Support

Professionals in either cancer or palliative care settings tended to have greater experience of caring for older adults. They had less experience providing end-of-life care to those aged 16-40 and fewer 'tools' or strategies to offer this younger population. Professionals found caring for young adults as they deteriorated both professionally and emotionally challenging and burdensome, as witness to young people prematurely reaching the end of their lives coupled with a weight of expectation to do more. The availability, accessibility and use of support for HCPs was variable and ranged from peer to professional support with a perception that experienced senior doctors were less likely to be in need. In contrast, nurses were perceived to be more likely to require and/or seek out support:

But there's always been this sort of demarcation that when they come to the – come to, "They're now incurable," they go somewhere else. And that 'somewhere else' is always nebulous. 'Someone else' looks after them 'somewhere else.' Do you know what I mean? … "Oh they go over there now." As I said earlier, the palliative team will look after them. And I *don't think any of us [Oncology CNS] have ever really gone to see what the palliative team do or see how much input they have. And is that a, is that a lack of professionalism or is that a survival mechanism for ourselves? And I have a feeling it's the latter. I have a feeling that it's very much a survival mechanism for ourselves because then we can just close that bit off and we can get on over here. And we'd like to know how they are, but we don't have to be the one that tells them. (Cohort 2 – HCP 16)*

Families Need Support

Families provided multiple types of support (practical, physical, emotional, financial) to the person with cancer to complement or supplement professional care:

Well I do as much for her as I can and I go out and do all her shopping. And if her husband is not around to pick the kids up from school, myself and my other daughter, we sort of take care of the kids. And also we've been taking them out as well because – and that upsets [name] more especially during the school holidays last week that they couldn't go anywhere. And she started saying, "I'm not a good mum." (Cohort 2 – Family 22)

The impending decline and death of a young family member was usually unanticipated and a situation that families have rarely experienced before. Family members generally had fewer appropriate skills to care for the person as their cancer progressed. Families expressed a wish for some form of access to information or training to care for their loved one appropriately. Looking back, bereaved families commented that their skills to deliver care at the end-of-life were limited and they would have liked access to some basic training and emotional support.

Language

The use of language by HCPs to describe an approach to care may not convey the same meaning to young adults with cancer and their families. For example, words such as hospice conjured up particular scenarios and carried ambiguity about the imminence of the end-of-life; such terms were often left unexplained, causing distress:

I do remember him [Consultant] saying, I can't really remember the conversation massively, but I do remember him keep saying, "Tumour, there's a tumour." And then I literally did have to say, "Hang on a minute, do you mean cancer?" and he said, "Yes, we've got to run more *tests and this, that and the other, but yes." But that's the only thing I remember really about it, if you know what I mean. (Cohort 2 – Patient 14)*

Financial Concerns

There were few participants for whom finance was not a concern. For those who were younger and still in education or training the burden tended to fall on their families. For those who were working, with loans, mortgages or dependents, the impact of cancer compromised their ability to support themselves and their families. Concerns were expressed about changes in lifestyle whereby the basics were prioritised. There was some confusion around entitlement to benefits or equivalent sources of financial support and limited access to tailored financial advice or guidance:

But you could do with somebody saying to you, in the first place, "You need somebody to help you to do this," you know what I mean, you need somebody who can guide you through the system. And I think the same applied with [name]. He'd think, "Oh well I've just got to fill this form in and I've got..." but actually filling those forms in is a damned hard job. (Cohort 2 – Family 23)

You haven't asked to be in that position [dying from cancer]. So I shouldn't have to go to work and think, 'Well I'll do a monotonous job just to pay the bills to only live another few months.' If I've only got a few more months to live, I'd rather spend it with my family, you know, having the time with them. (Cohort 2 – Patient 6)

Section 2 Realist explanations of our themes presented in the form of Context-Mechanisms-Outcome (CMO) configurations

We re-analysed our emergent themes using a realist logic of analysis. We attempted to identify mechanisms (generative causal processes) that are activated in the contexts we had found within the themes we uncovered. Our interview data were purely qualitative and so likely to be limited in the range of relevant data needed to build CMO configurations. To supplement these data, we deliberately drew on the extensive content expertise of the project team, workshops and where relevant, existing theories on needs of people living with cancer.

Details summarising the CMO configurations are presented in Table 3.

Table 3 CMO configurations, illustrative quotes and summary of our interpretations

Context-Mechanism- Outcome Configuration	Quote	Related theme
CMO 1 The diagnosis of cancer (context), changed the perception of control (mechanism) in young adults to cause distress, frustration and anger (outcomes).	*… like the feeling that I've got control over it, like complete control. For me that's extremely important. As soon as I lose that, I think I'd really struggle. And I need to, yes, feel as though I'm in the driving seat more or less. (Cohort 1 – participant 1)*	From the Loss of Control theme: The unexpected diagnosis of cancer disrupted everyday life and young adults often had to relinquish control and permit others to manage aspects of their life. The feeling of 'loss of control' was experienced throughout the diagnostic and treatment phases and seemed to increase when cure was not likely as participants experienced a loss of their anticipated future.
CMO 2 In the context of disease progression (context), young adults continued with normal activities as a coping strategy that offered distraction (mechanism) leading to a feeling of some kind of 'normality' (outcome). A poor prognosis and physical decline compromised the maintenance of a 'normal life'.	*"Yes but we're not going to do that" he [son] said, "We're just going to carry on as normal." And I thought actually he's right because carrying on normal makes it, it does make it more real. And more memorable … yes we do some lovely things, but it's just trying to keep everything as normal as possible really and just make the most of that time [Cohort 2 – family member – son did not participate]*	From the Maintenance of Normal Life theme: Young adults wanted to live as normal a life for as long as possible. Young adults and their families adjusted to a new normality, to accommodate the changes their disease progression created.
CMO 3 When there was trust between HCPs and a young adult (context), it was easier to introduce change or a new service (outcome) because a sense of abandonment (mechanism) or apprehension	*I think continuity is one thing that I'd put on a pedestal as being the most important, as a patient. It's horrible seeing different people and having to tell your story over and over and over again. Yet when you see somebody you know, and they know*	From the Continuity of Care theme: Young adults reported a high expectation and preference for continuity of care within and between services, which for them meant seeing the same HCPs whenever possible:

(mechanism) was less likely to occur. In contexts where continuity was provided (i.e. seeing the same HCP) trust developed (outcome) because of a sense of being known (mechanism).	*your story, they know whether you're well or you're not well. They know how your psychology works a little bit. So they know how to present things to you. That makes a huge difference to how you trust them, what your relationship is like and how you respond to them [HCP's] (Cohort 2 – Participant 7).*	
CMO 4 When a young person is dying because of cancer (context), HCPs find it challenging to talk about a poor prognosis or a shift in goal of treatment (outcome) because of their personal emotional discomfort of such discussions (mechanism).	*I find some of the younger patients it feels very unfair and I do reflect a lot on my own mortality and how I would cope (Cohort 2 – Participant 1 HCP)* *And it's a very stark contrast that [support] seems to be important for nurses but it's not seen to be important for doctors … as an individual, you don't talk because nobody wants you to talk about it, because you're the strong leader … Some doctors will just completely divorce themselves from it and will not engage in any shape or way with their patients … But I think you just sort of, you potentially just end up with, you know, increasingly tired and burnt out and disengaged doctors (Consultant Oncologist, HCP workshop 1)*	From the Professionals Need Support theme: The loss of a 'life partially lived' can be difficult for professionals, who feel a greater burden of sadness when young adults die HCP workshop participants felt support for them was often reactive rather than proactive with nurses more likely to receive support than doctors. A further distinction was recognised between junior and senior doctors:
CMO 5 When the way services are delivered for young adults does not fully recognise the additional needs of the family or care-givers (context), this leads to	*It is quite upsetting because, we actually felt abandoned, I felt abandoned.* *[Facilitator: By?]* *By just the whole system really. It was just, if you*	From the Families Need Support theme: Families often provided informal care for young adults within the home. Whilst they wanted to support their child or partner, caring created a

them feeling marginalised (mechanism) resulting in feelings such as abandonment and distress (outcomes).	*didn't ask, you wouldn't know (Family workshop 2- bereaved husband)* *And she [24 yr. old daughter] sort of became more and more sleepy and distant from us. But nobody would say to me, "This is what to look for. When she dies, this is what's going to happen. (Family workshop 3 – bereaved mother)*	further burden. Families felt insufficiently supported in this role.
CMO 6 When emotive language is used in palliative and end-of-life care (context), misunderstandings (mechanisms) can easily occur, leading to a range of different outcomes from encouraging hope through to despair (outcomes).	*So when my breast care nurses referred me to a hospice, I was like "Oh my god, that's horrendous, I don't want to do it". But, you know, it's been one of the best ever things. And I kept putting it off and saying, "I'm not ready for it, I'm not ready for it." And she [CNS] went, "Look, if you just make contact, then when you do need them, you can tap into them and they're quite good at financial advice". So I said, "Okay right let's do it". And actually they've been fantastic. Actually from just the level of sorting things out. (Cohort 2 – participant 19).*	From the Language theme: One example was the use of the term 'hospice'. When this was first raised with young adults their initial reaction was one of rejection as hospices were where older people went to die and young adults did not believe that they were at this stage:
CMO 7 Few young adults have thoughts about long term financial planning as they did not anticipate serious illness (context). This can lead to individuals and	*You haven't asked to be in that position. So I shouldn't have to go to work and think, 'Well I'll do a monotonous job just to pay the bills to only live another few months.' If I've only got a few more*	From Financial Concerns theme: Financial concerns and insecurity are almost always expressed by young adults with cancer.

families facing financial precariousness (mechanism). Access to tailored advice, whilst it may not solve financial concerns, may provide individuals and their families a range of ‘tools’ to better cope with their financial situation (outcome).	*months to live, I’d rather spend it with my family, you know, having the time with them. (Cohort 2 – Patient 6)* *But, yes, I think that’s the crappest thing, it’s not having – if you are single and I don’t have a rich family, you know, yes it’s just the whole worry of like affording things and knowing what kind of life you’re going to end up with if you give up work, especially when you’ve been used to a different kind of life. (Cohort 2 – Patient 19).*	

CMO: context, mechanism and outcome

Section 3 Revision of preliminary programme theory

Our re-analyses of the data enabled us to confirm, further develop and refine aspects of our preliminary programme theory – namely control, normality and family support. We were also able to add to our preliminary programme theory the concepts of continuity, professional support, language and financial support. Below we summarise important aspects of our better refined programme theory.

Age specific issues

We now understand that *for those aged 16-40 there are specific differences between the end-of-life care experience and preferences.* However, *rather than being wholly defined by age, the stages in a young person's life course may be a better way to approach, understand and support these differences.*

Maintenance of control and sense of normality

Our data underpin these concepts within our preliminary programme theory. We have learned that *young adults with cancer need support to put strategies in place to retain control and live as normally as possible whist providing a space to discuss and plan for their shortened future.*

Families of younger people with cancer

We found that *the family often are not appropriately equipped to provide the level of care and support that they want to provide during the last year of life of the young adult with cancer and lack the means to be 'skilled-up' for this role.*

Healthcare professionals

We found that *healthcare professionals lack age (16-40-year-old) life course-specific knowledge to develop strategies to support patients in their last year of life and their families.*

Discussion

In this study, we used a realist evaluation approach to gain a deeper understanding of the particular contexts that may be specific to the experiences of young adults aged 16-40 years with cancer as they approached their end-of-life. We re-analysed our initial seven themes into seven CMO configurations that explained the specific needs of the end-of-life experiences of young adults with cancer. The implications of these specific needs are set out below and compared and contrasted with the existing literature.

Life course and not age matters

We found that within this group, end-of-life experiences and preferences cannot be neatly isolated into the two age ranges we studied (16-24 years and 25-40 years). A better way of approaching, understanding and supporting young adults may be to consider where they are in their life course, as there may be more in common, than different, between those with similar life course experiences, for example being in education, maintaining a career, having children or caring responsibilities. The usefulness of taking such an approach is also found in the wider literature on end-of-life care for young adults and so reinforces this finding.(18) Adolescence and young adulthood is a developmental stage when individuals shape their identities, gain autonomy, make career choices and develop intimate relationships. A cancer diagnosis at this stage is “off-time” during the normative life cycle: life is interrupted, developmental tasks and identity formation are challenged and few peers will share their cancer experience.(18) In common with Soanes and Gibson we found that participants across this age range reported a desire to maintain these aspects of their life, as well as their identity for example, as a student, a professional, or parent, in part to maintain a sense of normality and control.(19)

Giving young people the chance to have control and to feel normal

We found, perhaps unsurprisingly, the pivotal role of HCPs in supporting young adults with cancer. However, we were able to identify that an important ‘block’ to the support provided comes from the emotional discomfort felt by HCPs when discussing aspects of care specifically with young adults – such as discussions about prognosis. This is important as a cancer diagnosis creates great uncertainty and the knowledge that there will not be a cure creates a dissonance between the life that was expected and the reality of a life that will be significantly shorter than expected. For emerging adults and early independent adults, as disease progresses, dissonance is also present as their independence is compromised with an increasing and unanticipated dependence on others. This can affect their ability to attend school, college or work as well as taking part in family or social activities or fulfilling caring duties for others e.g. looking after young children. Adaptation is a mechanism through which there is a recognition of what can no longer be achieved due to disease progression.(20) An adapted normality can be achieved together with a sense of control, allowing for realistic goal setting.(21) Advance care planning could facilitate this adaptation. However, few participants in our study reported having had conversations about their options or the care they wanted to receive. Some HCPs avoided such conversations because of the emotional burden to themselves, not wishing to challenge either hope or a young person’s possible denial about their situation. This might be an example of what Bell et al refer to ‘as social constraint’, i.e. words and actions that inhibit end-of-life discussions.(22) A further notable finding from the data indicates that all parties appear to wait for another to raise the topic of end-of-life. The ‘window of opportunity’ (17) often fails to appear, thus in some cases the topic is avoided. This has the potential to delay adaptation and limit the time available for professional support, which could help young adults plan and make as much as possible of remaining time.(23) For those with dependents, particularly young children, delaying adaptation could impact on their roles as parents, delaying the opportunity to prepare and create memories for themselves and their families.(21) When end-of-life was addressed, this tended to be when health had deteriorated, and that window of opportunity, albeit late, facilitated opportunities to discuss the future, end-of-life care and to make plans.

Families and carers matter even more

Data from family members came from two perspectives – both before and into bereavement. Many family members became informal caregivers. We found increased dependence on family members whether emotionally, physically, financially or for support with housing. The level of independence varied between the two age groups with those aged 16-24 more likely to be living in the parental home, still in education or receiving training and moving towards becoming independent from their family. Those aged 25-40 were more likely to have been independent adults for longer. In common with Knox et al, we also found that when thrust back into dependent relationships with parents, left behind by peers, whom they perceived to be moving forward with their own life goals, young adults could feel isolated.(24) The financial burden of cancer is widespread, but for those at the younger end of the age group who were still in education or living at home, the burden fell more heavily on their family. For those with greater independence and who relied on their income from employment, a cancer diagnosis compromised their ability to work and maintain their lifestyle. It is likely that older people with cancer, particularly those who have retired with an income to cover their regular expenses may not face such financial extremes. Mohammed et al. refer to caregivers ‘taking charge’, thrust into a role for which they often felt ill-prepared.(25) In our study, lack of understanding of the clinical situation due to confidentiality, a lack of practical or technical knowledge or skills and poor information from HCPs, themselves often reluctant to undertake end-of-life discussions, were some of the contextual influences contributing to feeling ill-prepared, abandoned or distressed.

The burdens for healthcare professionals

Professionals reported difficulty addressing the needs of both the person with cancer and their family as often they had different expectations. Professionals were aware that providing bereavement support to a family was difficult if they had not built a relationship with them in the limited time available. This is mirrored by our finding that continuity mattered much more to young adults. Managing complex family dynamics was challenging for HCPs and strategies to do this were often not addressed. Sometimes HCPs did not want to ‘open a can of worms’ by involving the family as they were aware that they would have to consider extra care needs, not viewed as part of their role. This was a strategy used by HCPs to manage their workload and families were not told that it was acceptable to ask for help and support. Professionals preferred to maintain and share optimism with the family, maintaining hope, all of which helped to reduce the emotional discomfort they would otherwise feel. So, talking openly about the death of the person with cancer was rarely pursued. Beerbower et al. refer to ‘a broken system of communication’ that can lead to conflict, where there has been no disclosure of prognosis, or where disclosure has for some family members only been partial, or come much too late.(18, 26) Educating, enabling and supporting caregivers can thus be complex and challenging, reinforcing the need for early and developmentally appropriate communication.

Professionals often have less exposure to and experience of providing end-of-life care for young adults. They are likely to be similar in age to the person with cancer, their family or friends, enhancing the emotional difficulties of working with this population. Whilst in palliative care, end-of-life might be ‘normal’, caring for those aged 16-40 who are dying will not be normal nor will facing the loss of lives partially lived. The avoidance by HCPs of engaging in the challenging discussions and activities we have already listed is understandable. But Wiener et al. point out that HCPs need to reflect and be aware of the emotional effect that younger patients have upon them and whether the support they offer is relevant and enabling of this population to continue to live normally for as long as possible.(27) Clark et al. have suggested that providing a developmentally-appropriate approach to care that includes advance decision making is thus essential.(28) To enable HCPs to meet the needs of the end-of-life care of young adults, formal support is needed. However, the formal support for HCPs in their professional roles varied in availability, access and was used differently. There was a distinction between doctors and nurses. Participants in our study suggested that the emotional burden received greater recognition in the nursing profession whereas for senior doctors there was little or no provision of support and an expectation that they would not show the emotional effect of their work. There were also issues about having the time to access support, associated costs and the lack of visibility and advocacy from senior HCPs for accessing support. In addition, support was not integrated into training or ongoing professional practice and for some senior HCPs it may have been regarded as compromising their role or authority.(29) Self-care in the palliative care workforce is known to be essential, yet rarely is education or training available.(30, 31)

We would agree with Knox et al that palliative care services should consider prioritising resources to support self-care practice, to promote the health and well-being of HCPs.(18)

Strengths, limitations and future research directions

Although our study is unusual for the extensive data collected from young adults facing end-of-life and their triangulation with family and HCPs, recruitment of two groups of patients was unsatisfactory. Young adults with haematological malignancies were rarely invited to participate despite these being a commoner diagnosis in this population. This may be because those with haematological diagnoses continue to be offered and agree to receive ‘curative’ treatments.(32) When such curative options had been exhausted our participants were often ’actively dying’ and too ill to participate in this study. Another under-represented group were those aged 16-24. Professionals suggested that whilst clinical teams identified young adults meeting the study eligibility criteria, the challenges of communicating that ‘cure was not likely’ may have impacted on their reluctance to introduce the study. Our original plan to undertake two interviews with participants failed: often patients were just too unwell for a second interview. We cannot be certain that the data presented in this paper wholly reflects the experiences of these two populations, therefore further research is needed to explore the needs of those often described as ‘hard to reach’, those with haematological cancers and those aged 16-24 years.

Conclusion

We identified challenges with the way current end-of-life care is delivered to young adults with cancer. Using this evidence, recommendations to improve care can now be developed.

References

1. Bleyer A, Ferrari A, Whelan J, Barr RD. Global assessment of cancer incidence and survival in adolescents and young adults. Pediatr Blood Cancer. 2017;64(9).

2. What are the top causes of death by age and gender? [Internet]. 2012 [cited 02 August 2016]. Available from: http://webarchive.nationalarchives.gov.uk/20160105221750/http://www.ons.gov.uk/ons/rel/vsob1/mortality-statistics--deaths-registered-in-england-and-wales--series-dr-/2012/sty-causes-of-death.html.

3. Globocan 2012: Estimated Cancer Incidence, Mortality and Prevalence Worldwide in 2012 [Internet]. 2012 [cited 02 August 2017]. Available from: http://globocan.iarc.fr/Pages/fact_sheets_population.aspx.

4. Ngwenya N, Kenten C, Jones L, Gibson F, Pearce S, Flatley M, et al. Experiences and Preferences for End-of-Life Care for Young Adults with Cancer and Their Informal Carers: A Narrative Synthesis. J Adolesc Young Adult Oncol. 2017;6(2):200-12.

5. Bleyer A, Barr R, Ries L, Whelan J, Ferrari A, editors. Cancer in Adolescents and Young Adults. 2nd ed. Switzerland: Springer; 2017.

6. Grinyer A. Palliative and End of Life Care for Children and Young People: Home, Hospice, Hospital. Oxford: Wiley; 2012.

7. Grinyer A, Thomas C. The importance of place of death in young adults with cancer. . Mortality. 2004;9(9):114-31.

8. Kars MC, Grypdonck MH, de Bock LC, van Delden JJ. The parents' ability to attend to the "voice of their child" with incurable cancer during the palliative phase. Health Psychol. 2015;34(4):446-52.

9. Kars MC, Grypdonck MH, de Korte-Verhoef MC, Kamps WA, Meijer-van den Bergh EM, Verkerk MA, et al. Parental experience at the end-of-life in children with cancer: 'preservation' and 'letting go' in relation to loss. Support Care Cancer. 2011;19(1):27-35.

10. Baker JN, Hinds PS, Spunt SL, Barfield RC, Allen C, Powell BC, et al. Integration of palliative care practices into the ongoing care of children with cancer: individualized care planning and coordination. Pediatr Clin North Am. 2008;55(1):223-50, xii.

11. Grinyer A, Barbarachild Z. Teenage and young adult palliative and end of life care service evaluation. London; 2011.

12. Pawson R, Tilley N. Realistic evaluation. London: Sage Publications Ltd; 1997. 235 ; 23 cm. p.

13. The Rameses II Project. “Theory” in realist evaluation 2017 [Available from: http://ramesesproject.org/pgm-download_media.php?name=RAMESES_II_Theory_in_realist_evaluation.pdf.

14. The Rameses II Project. Developing Realist Programme Theories 2017 [Available from: http://ramesesproject.org/pgm-download_media.php?name=RAMESES_II_Developing_realist_programme_theories.pdf.

15. Wong G, Westhorp G, Manzano A, Greenhalgh J, Jagosh J, Greenhalgh T. RAMESES II reporting standards for realist evaluations. BMC Med. 2016;14(1):96.

16. Richards L, Qualitative S, Research. Using NVivo in qualitative research. London: Sage; 1999. ix, 218 pages : illustrations ; 25 cm. + 1 CD-ROM (4 3/4 in.) p.

17. Charmaz K. Grounded Theory: objectivist and constructivist methods In: Denzin NK, Lincoln YS, editors. Handbook of qualitative research. California: Sage Publications Ltd; 2000. p. 1065 ; 26 cm.

18. Beerbower E, Winters D, Kondrat D. Bio-psycho-social-spiritual needs of adolescents and young adults with life-threatening illnesses: Implications for social work practice. Soc Work Health Care. 2018;57(4):250-66.

19. Soanes L, Gibson F. Protecting an adult identity: A grounded theory of supportive care for young adults recently diagnosed with cancer. Int J Nurs Stud. 2018;81:40-8.

20. Livneh H. Psychosocial Adaptation to Cancer: The Role of Coping Strategies. Journal of Rehabilitation. 2000;66(6):40-50.

21. Nissim R, Rennie D, Fleming S, Hales S, Gagliese L, Rodin G. Goals set in the land of the living/dying: a longitudinal study of patients living with advanced cancer. Death Stud. 2012;36(4):360-90.

22. Bell CJ, Zimet GD, Hinds PS, Broome ME, McDaniel AM, Mays RM, et al. Refinement of a Conceptual Model for Adolescent Readiness to Engage in End-of-Life Discussions. Cancer Nurs. 2018;41(2):E21-E39.

23. Llewellyn H, Neerkin J, Thorne L, Wilson E, Jones L, Sampson EL, et al. Social and structural conditions for the avoidance of advance care planning in neuro-oncology: a qualitative study. BMJ Open. 2018;8(1):e019057.

24. Knox MK, Hales S, Nissim R, Jung J, Lo C, Zimmermann C, et al. Lost and stranded: the experience of younger adults with advanced cancer. Support Care Cancer. 2017;25(2):399-407.

25. Mohammed S, Swami N, Pope A, Rodin G, Hannon B, Nissim R, et al. "I didn't want to be in charge and yet I was": Bereaved caregivers' accounts of providing home care for family members with advanced cancer. Psychooncology. 2018;27(4):1229-36.

26. Bell CJ, Skiles J, Pradhan K, Champion VL. End-of-life experiences in adolescents dying with cancer. Support Care Cancer. 2010;18(7):827-35.

27. Wiener L, Weaver MS, Bell CJ, Sansom-Daly UM. Threading the cloak: palliative care education for care providers of adolescents and young adults with cancer. Clin Oncol Adolesc Young Adults. 2015;5:1-18.

28. Clark JK, Fasciano K. Young adult palliative care: challenges and opportunities. Am J Hosp Palliat Care. 2015;32(1):101-11.

29. Bradford NK, Greenslade R, Edwards RM, Orford R, Roach J, Henney R. Educational Needs of Health Professionals Caring for Adolescents and Young Adults with Cancer. J Adolesc Young Adult Oncol. 2018.

30. Mills J, Wand T, Fraser JA. Self-Care in Palliative Care Nursing and Medical Professionals: A Cross-Sectional Survey. J Palliat Med. 2017;20(6):625-30.

31. Mills J, Wand T, Fraser JA. Examining self-care, self-compassion and compassion for others: a cross-sectional survey of palliative care nurses and doctors. Int J Palliat Nurs. 2018;24(1):4-11.

32. Odejide OO, Cronin AM, Earle CC, Tulsky JA, Abel GA. Why are patients with blood cancers more likely to die without hospice? Cancer. 2017;123(17):3377-84.

Legends for figures

Figure 1. Phases of research process. Phases ii-iv are reported here.

Figure 2. Analysis process

Figure 1. Phases of research process. Phases ii-iv are reported here

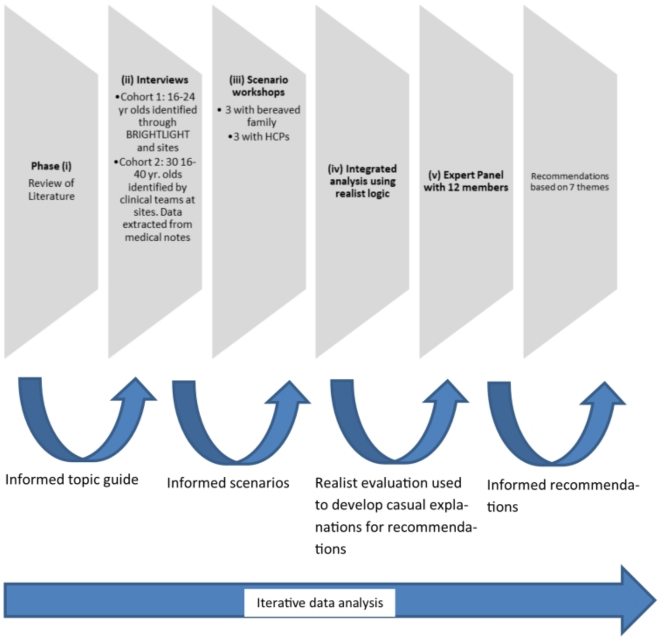

Figure 1 Phases of research processes Phases ii-iv are reported here
Figure 2 Analysis process

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University College London Hospitals NHS
NHS Foundation Trust

Title of project: When cure is not likely: What do young adults with cancer and their families need and how can it best be delivered? A BRIGHTLIGHT companion study

PROTOCOL
Version 2.0 19th December 2013

UCL

Protocol Authorisation

PRINCIPAL INVESTIGATOR

Print name ………………………………………………………………..

Signature ………………………………………………………………..

Date ………………………………………………………………..

Amendment History

Version	Date	Author	Application reference	Notes

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Lay Abstract

Background

This study will help increase our knowledge to understand better the needs of 16-40 year olds (“young adults”) when cure from cancer becomes unlikely. The younger members of this group are on the borders of paediatric care and as cancer is relatively uncommon in this age group, patients are less often encountered in adult cancer and palliative care services. Overall cancer is relatively uncommon in people in early to mid-adulthood. Nevertheless, one quarter of deaths in 16-40 year olds are due to cancer. The impact of incurable cancer on patients and families is strongly influenced by age but for adults in early to mid-life, very little is known about their experiences as death approaches or how care is best delivered. Professionals consistently acknowledge many specific challenges of managing end of life care in this age group.

From this study we aim to understand;

a) The most important parts of care in the last year of life for people with cancer aged 16-40 years.
b) Whether differences exist between the experiences of people with cancer who are aged 16-24 and those aged 25-40 years.
c) How young adults and their families can be supported in the last year of life to achieve their preferences for care.
d) The challenges that exist for health and social care professionals providing care.

Methods

There are five distinct parts to our research.

I. **Analysing available information:** we will look at all available literature and information about end of life care in young adults including the information that is now collected routinely in the NHS about preferences and place of death.
II. **Interviews with patients, families and professionals**. We will work closely with health care professionals from four study sites (University College London Hospitals, Southampton University Hospitals, Leeds Teaching Hospitals, and St. Joseph’s Hospice) to identify patients for interview about their care when cure is no longer likely. Fifteen 16-24 year olds taking part in the BRIGHTLIGHT study (a study already underway in the UK) and thirty 16-40 year olds from the four study sites will be invited to participate. We will ask them to propose a family member or carer and their key worker to be interviewed as well.
III. **Workshops with patients and professionals.** We will conduct nine workshops for interactive discussion and to increase our understanding of the range of perspectives, opinions and experience. Three workshops will be with family members and carers, three with professionals and three bringing together family members and carers and professionals.
IV. **Hold professional panels.** We will present our findings and recommendations to a panel of professional experts for further refinement.
V. **Make recommendations for practice.** We will have a good understanding of the core components and pathways of end of life care for young adults and make recommendations for practice and further evaluation to assist policy makers, commissioners and other stakeholders.

How the results of this study will be used

The findings will add evidence to inform national standards, pathways of care and core competencies for training staff. We will identify areas for change or further exploration. We will make public our findings widely to both lay and professional audiences.

1. Purpose of the study

This research will illuminate the core issues affecting end of life care[1] in young adults with cancer (aged 16-40 years), gathering evidence from the perspectives of the young people themselves, their families and the multidisciplinary team.

Using a national, multiple method realistic evaluation, we shall use an iterative approach guided by the MRC framework for evaluating complex interventions. [1,2] The aim is to define, describe and understand the core components for excellent practice in the delivery of end of life care for young people with cancer, to inform policy and practice and to set priorities for further evaluation studies.

In the United Kingdom (UK), health policy on specialist cancer services has bracketed young adults up to 24 years with teenagers (teenagers and young adults, TYA). [3] In the United States, Canada and Australia, strategies for improving cancer outcomes have most often focussed on 15-40 year olds ('adolescents and young adults', 'AYA'). [4] Notably, Douglas House, a unique hospice for young adults in Oxford serves an age range of 16-35 years. [5] This age group has attracted little attention in other European countries where there has been less consistency in the age range studied. We aim to explore comparisons between those aged 16-24 years and those aged 25-40 years to highlight key issues and differences that may be influenced by age.

In devising this research study, our underlying theories are:

1. That end of life care for young adults with cancer aged 16-40 years could be improved by increased understanding of (a) current care pathways for people with different types of cancer (b) the effects of age (above and below 25 years) (c) the need for accurate information (d) how active participation by young adults in decision making can impact on current and future care (e) the importance of respect for individual autonomy and family interactions.

2. That outcomes would be improved by an approach to end of life care that is (a) more aware of individual patient need and autonomy (b) supports professionals to recognise and respond to patient need (c) takes account of family interactions and relationships with patient and staff (d) enables patients to receive active and palliative treatments in a place of their choice to achieve their preferences for end of life care.

Thus, our objectives are to understand:

a) The core components in the pathways of care in the last year of life for people with cancer aged 16-40 years.
b) Any differences between the experiences of people with cancer from the age ranges 16-24 and 25-40 years.
c) How young adults and their families can be supported in the last year of life to achieve their preferences for care.
d) The challenges that exist for health and social care professionals providing care.

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[1] End of life care is defined as 'care that helps all those with advanced, progressive, incurable illness to live as well as possible until they die'. [6]

2. Background

One quarter of deaths in 16-40 year olds are related to cancer.[7] Overall survival rates have improved less than those of younger children and older adults.[8] Despite descriptions of the specific needs of teenagers and young adults for specialist cancer care, there is a dearth of empirical research, policy and expert practice related to their End of Life care. This is also true for young adults up to 40 years.

There are gaps in policy for this age group. Better care: Better Lives [9] makes no distinction between the needs of children and teenagers and young adults and deals exclusively with children's palliative services. Similarly the End of Life Care Strategy, Promoting High Quality Care For All Adults At The End Of Life makes no specific reference to young adults.[10]

Published literature about End of Life for teenagers and young adults has been confined to summaries of good practice or, where studies have been undertaken, data has been sourced from parents rather than young people themselves.[11-13] Others have undertaken retrospective analysis of medical notes [14] or produced comment and review papers.[15-18] Notably, just one study interviewed young people and explored their views on decision making as End of Life approached.[19]

Equally, work on the older young adult cancer population is limited. The few studies that have collected data from adults of all ages, either through interviews or questionnaires (EORTC QLQ-C30) have identified differences between ages at end of life including varying preferences for active treatment and greater symptom burden or lower quality of life.[20-22]

For those aged below 40 years, boundaries between curative and palliative treatment are often blurred; decisions may be influenced by cancer type, age and family circumstances. There is a lack of standardized models, approaches and communication aids for this age group; the management of symptoms and psychosocial concerns may be neglected if the challenges to communication are not overcome. Avoidance and delaying of discussions about End of Life decisions are common resulting in consequences for the quality of care.[15]

The challenges in delivering care to young people are as relevant to the delivery of end of life care as to intensive curative chemotherapy.[23-24] National policy in England and Wales directs that TYA have access to specialist services to meet specific challenges faced by young people in accessing services responsive to needs associated with the physical, educational, psychological and social developmental stages that are disrupted by the diagnosis and treatment of cancer.[4] This policy is being evaluated in the BRIGHTLIGHT study, (NIHR RP-PG-1209-10013; Appendix 3, study synopsis).

BRIGHTLIGHT will provide: a cohort of participants of all those aged 13-24 diagnosed with cancer in England in one year; the contexts and pathways of cancer care for young adults in England; sources of contributors to workshops and expert panels; prospectively collected patient outcome data and extensive analysis of the national cancer and end of life data sets. This takes into account information already available in the national cancer data repository (NCDR) and from the National End of Life Care intelligence network (Nend of lifeCIN) for those aged 16-40 years. BRIGHTLIGHT and this proposal are closely linked to the remit of the National Cancer Research Institute Teenage and Young Adult Clinical Studies Group.[25]

This study will provide the much-needed evidence to underpin quality of care and the enablers that will assist in addressing current barriers:

	Barriers to excellent end of life care	Enablers of excellent end of life care

We can influence	Evidence base: Poor understanding of (a) disease trajectory; (b) unmet health and social care needs; (c) variations between cancers; (d) variations between age, groups, gender, ethnicity Service factors: Support needs of staff , patients and families in decision making Attitudes and barriers to care of young people: structural; cultural; financial. Individual factors: personal and disease specific, education Costs of care	Data from BRIGHTLIGHT, NCDR and Nend of lifeIN Data from workshops and interviews Training and support health and social care professionals Understanding of the costs of components of end of life care components
What will influence our project	Recognition of last year of life and end stage disease Interaction between active and palliative treatments Family dynamics Attitudes of health care professionals National factors in healthcare environment; costs of care (perceived and actual), commissioning Regional variation in policy, service configuration and provision; cultural and economic factors Training and support needs of health and social care professionals	Integrated care pathways Strategic documents; DH and voluntary sector pressures; new service providers; public opinion; epidemiological trends Assessing transferability of intervention in regional sites Engagement of clinical champions; multi-disciplinary team approach to care Training and support for health and social care professionals

3. Study Design and Methods

Research will be undertaken in 5 discrete but inter-related phases.

(i) Preparatory phase;
(ii) Interviews with young adults, families and professionals;
(iii) Scenario workshops with families and professionals;
(iv) Analysis and interpretation of these data including synthesis with a literature review and quantitative data available through NCDR, Nend of lifeCIN and BRIGHTLIGHT then refinement through discussion with a panel of experts;
(v) Development of recommendations for practice and further evaluation.

See Appendix 1 for diagram of these phases.

(i) Preparatory phase

This will be undertaken at UCLH and includes a detailed synthesis of the literature relevant to end of life for young adults with particular emphasis on identification and understanding of the mechanisms potentially causing the desired outcomes. Additionally, patterns of care described by National Cancer Data Repository and National End of Life Care Intelligence Network will be used to give further characterisation by definition of tumour types responsible for deaths, place of death and variations within our age range. This phase will inform the semi-structured realist interviews with young adults and scenario development.

(ii) Interviews

Several groups including patients, families and professionals will be interviewed as described below. Four sites will be involved in this part of the study.

We will conduct interviews with 45 young adults aged 16-40 years of age, purposively sampled to reflect a range of diagnoses (relevant groupings include leukaemia, lymphoma and solid tumours including brain, testis, sarcoma, carcinomas). Recruitment will be from the two sources outlined below (Appendix 2 illustrates recruitment, consent and data collection processes).

Sample 1 will be 15 teenagers and young adults aged 16-24 years participating in the BRIGHTLIGHT[2] cohort who will ‘self-identify’ through responses to trigger questions in the BRIGHTLIGHT survey indicating that a) no more treatment is possible, b) they have been offered/received care from the symptom control team or palliative care team or c) giving a response to the question asking what they had been told about their cancer suggesting that they are aware that cure is no longer likely. The BRIGHTLIGHT Senior Research Manager (SRM) or Cohort Manager (CM) will confirm with the young person’s healthcare team that they are receiving end-of-life care (if this was not confirmed on the pre-survey check) and make sure that it is suitable to approach them to take part in the study. When this is assured they will call the young person, briefly describe the study and gain verbal consent to forward the information sheet. After approximately a week, the young person will be contacted via an appropriate means (e.g. ‘phone, text) by the same person (SRM or CM) to see if they have received the information sheet and asked if they would like to take part in the study. For those who agree they will be asked for verbal consent to pass their contact details onto the RA who will then contact the young person.

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[2] See www.brightlightstudy.com for details of BRIGHTLIGHT and appendix 3 for a synopsis.

If a young person verbally consents to participate the RA will arrange a time and place to meet them for the interview. On the day before the interview the RA will call to confirm the young person would still like to participate; if they do then the RA will visit the young person as planned but before commencing the interview, s/he will confirm the young person understands what they are agreeing to and get written consent. For a detailed description of this recruitment process, refer to Appendix 4.

After a first interview, this group will be invited to participate in a second interview between 2 and 4 months later. At the time when the young person is recruited, they will be asked to nominate a key worker who will be in charge of providing support during the study. This key worker will be in contact with the researchers and will follow up with the young person after the interview to see if they need additional support.

Sample 2 will be interviews to include thirty patients aged 16-40 years. Recruitment will be facilitated by clinical staff at six study sites: University College London Hospitals, Southampton University Hospitals, Leeds Teaching Hospitals, St Gemma's Hospice, Wheatfields Hospice and St. Joseph's Hospice. These research sites were selected because they all have established palliative care teams and services, they actively participate in research, and the staff are skilled at recruiting patients in End of Life for research.

A poster will be displayed in appropriate spaces with contact details of the site PI and potential participants asked to talk to their clinician if they are interested. Once members of the clinical team have identified potential participants, they will make sure their details can be shared with the researchers. If the participant agrees, the researchers will contact them with more information about the study. Whenever possible, this will be done through a face to face conversation about the study, but if this is not possible, the information will be sent via post and questions will be answered over the telephone. The participants will then be given time to look at the information sheets and ask questions about the study. If the participant agrees to take part in the study, they will be asked to sign a consent form.

The young people in sample 2 will be asked to identify a family member, and a professional directly involved in their care such as the key worker who can be approached to be interviewed in addition (total 90 interviews). The young people will be asked to check with the family member to see if they agree that their details be passed to the researchers so that they can contact them with more information about the study. If the family member agrees, the researchers will contact them to talk about the study and provide them with the information sheet. The researchers will also approach the nominated healthcare professionals to provide them with more information about the study and to see if they are interested in taking part.

This second sample will allow targeting of patients identified by their key workers as being directly aware that cure is no longer likely and currently in the end of life phase. Ten will be aged 16-24, and 20 between 25-40 years. The sample size and distribution has been selected to add additional interviews to complement sample 1, to reflect the clinical heterogeneity present across this age range and in anticipation of saturation of themes during analysis (refer to Appendix 5 for the sampling matrix).

Using an investigator designed template (see Appendix 6), medical notes of interviewees will be reviewed to analyse written communication and documentation about discussions related to end of life in order to seek illustrations of best practice. Records of key discussions will be sought including communication between health professionals; record of advance care planning, do not resuscitate orders; communication about preferred place of death; insight of patient and family; information given and received; and evidence to show if limits of desired levels of information had been elicited. We will pilot the review of the notes at the beginning of the period of data collection and adjust the template to suit the content found in the medical notes.

All semi-structured realist interviews will be conducted by a researcher experienced in working with young adults and discussing sensitive issues. It is anticipated they will last for approximately one hour, will be digitally recorded, and transcribed verbatim prior to qualitative analysis of transcripts. Interviews will occur in the place of choice of participants, usually during clinic visits or in their own homes.

(iii) Workshops

Scenario development: end of life trajectories for young adults with cancer will be derived from an initial scenario development workshop attended by palliative care consultants, palliative care and cancer clinical nurse specialists, allied health professionals, oncologists and haematologists. Findings from the literature review, analysis of National Cancer Data Repository and National End of Life Care Intelligence Network data and emerging findings from the interview data will be presented. Scenarios will be co-constructed to precipitate discussion around key influencing variants such as diagnosis, different ages within the range being studied, symptom constellations and patterns of information disclosure and communication (for examples refer to Appendix 7).

Nine scenario workshops: A series of three workshops will be held in the study sites used in (ii) above using the scenarios to encourage interactive discussions and generate new thinking and potential solutions to problems raised. The use of different sites will support comparisons across contexts and address issues of generalisability and transferability of findings across different UK settings. Workshops at each site will be held with:

1. Families and carers of young adults with cancer; patient representatives and regional stakeholder organisation representatives (workshop maximum size 15)
2. All members of the multiprofessional team involved in delivery of treatment for cancer and end of life care (workshop maximum size 15)
3. Mixed participants representing groups (1) and (2) (workshop maximum size 20)

The family members and healthcare professionals will be recruited using two routes. First, from the participants who were interviewed as part of sample 2. The consent forms for the interviews explained in (ii) will include a section where the family members and healthcare professionals can indicate if they would like to be contacted to participate in the workshops. Second, additional family members and healthcare professionals will be recruited in the study sites by the researchers and members of the clinical team. Close contact with the clinical team will facilitate recruitment.

Consensus will not be sought, rather perspectives, opinions and experiences elicited allowing scenario ‘mapping’. This will be done in small groups using mapping aids to identify key ideas and hypotheses arising on how end of life care could be improved.

All workshops will be moderated by members of the research team who have experience of these methods as well as end of life/cancer care. Workshop teams will be developed to ensure consistency across this aspect of data collection.

(iv) Expert panel review

A panel of experts will be convened to include palliative care professionals, general practitioners, oncologists, haematologists, allied health professionals, policy makers, ethicists, charities and commissioners. Participants will not have been involved in previous workshops or interviews. Professionals will be identified by previous participants and members of the

research team. Professional organisations such as the NCRI Palliative and Supportive Care Clinical Studies Group will be consulted. These will be purposively selected based on their experience working with young people who are receiving end of life care, knowledge of policy issues and agree to participate.

A distillation of knowledge and understanding gained from realist interviews and workshops combined with quantitative data, will be presented to the expert panel describing ideal pathways with an explanatory account of key components of care. The panel will be asked to comment on areas of agreement and disagreement, provide alternative or additive explanations and to test and retest experientially the fit of mechanisms to potentially achievable outcomes as described in the pathways.

(v) Development of recommendations

The analysis and interpretation by an expert panel will lead to the writing and refining of an overarching explanatory account for end of life care for young adults with cancer. We shall present data to inform our objectives stated in section 1.

Emerging national standards, pathways of care and core competencies are anticipated and will be considered within the document. We shall circulate our draft to participants in the realist interviews and workshops, policy makers, commissioners and other stakeholders and take account of their comments in producing a final version. Identification of areas for potential intervention will be included and recommendations for future empirical evaluation and testing of the effect on outcomes will be suggested. Such empirical work would be the subject of the next phase in testing the feasibility and acceptability of the core components of delivering improved end of life care in a pilot observational study or exploratory randomised trial in the future. This would include consideration of the economic aspects of these approaches to care.

4. Data analysis

Methodological approach

We will use realistic evaluation, derived from critical realism. Its strengths are an emphasis on understanding the causal mechanisms which generate outcomes, consideration of context and a desire to improve practice and service delivery.[26] It supports a mixed method, iterative approach to capture multidimensional aspects of the evaluation of end of life care, which can be viewed as a complex, multi-component intervention. Realist research explores the links between context, mechanism and potential outcome. It increases our understanding of 'what works, for whom, in what circumstances, in what respects and why?' It seeks to penetrate beneath the observable inputs and outputs of an intervention. We shall initially untangle the influence of context in the care of young adults in age groups 16-24 and 25-40 years, with a range of cancer diagnoses, experiences and expectations of care. We shall use workshops to generate hypotheses on mechanisms by which care in the last year of life for people aged 16-40 years might be improved. Using qualitative data and quantitative data we shall assess which components of an intervention or approach to end of life care might lead to improvements in patient and family centred outcomes.

Analysis of interviews

Sample 1: Transcripts of the interviews will be analysed using a grounded theory approach. This provides a systematic and inductive approach for the collection of data, sampling and the building of theoretical frameworks.[27] Analysis will occur simultaneously with data collection. After reading and re-reading the transcripts, memoing and selective focused coding, constant comparison between codes will take place leading to development of categories. Software such as Atlas.Ti will support this. The second interviews facilitated by theoretical sampling will ensure completion of any conceptual gaps in the emerging theoretical framework with an understanding of individual experiences over time.

Sample 2: Grounded theory methods of analysis will also be used. The development of codes and constant comparison of codes in the development of categories will occur across the sources of data for each young adult. For example data will be triangulated using the interview data from the young person, their family member, nominated health professional and analysis of the medical notes. When a conceptual understanding has been developed for each case, analysis will occur across the sources of data as a whole.

Analysis of workshops

Detailed field notes will be taken, memos and post-it notes collected and workshop discussions will be audio-taped and transcribed verbatim as discussion around scenarios is as crucial as the ‘mapping’.[28] Data will be entered into Atlas. ti. The research team will create a preliminary analysis from initial observations of the scenario maps developed and the transcripts of discussions from workshops (1) and (2) to generate a series of hypotheses of how end of life care can be improved. This initial analysis will be used to inform discussions in workshop (3).

After the data from the workshops is analysed, a short report will be distributed among the participants in order to share the findings as near to the time as data collection as possible.

Analysis for expert panel review

Analysis of data collected in an iterative process, a key aspect of realist methods, will be the focus. Preliminary thematic summaries of findings from the interviews and workshops will be combined with emerging quantitative data. Hypotheses on mechanisms of how end of life care could be improved will be carefully defined and prioritised. These will be refined further through discussion within the research team and with a panel of experts. This phase will allow a reconsideration of understanding of the interrelationships between the context and mechanisms generated by the experience of end of life care derived from all aspects of data collection, testing assumptions and exploring further remaining uncertainties.

5. Ethical considerations

We recognise that this is a sensitive area of research and we will be working with vulnerable participants. We anticipate the fact that the questions included in our interview could pose potential emotional and psychological burden for those involved. We are sensitive to this fact. The clinical members of our research team have significant experience in this area that we can draw upon. In addition we will make use of the now growing body of evidence of published studies on this topic.[29,30]

Therefore, we have taken the following measures in order to minimise the risks and burden for the research participants. The researchers to be appointed will have extensive experience of gaining consent and interviewing vulnerable participants. The researchers will ensure the protection and well-being of the participants throughout the entire duration of the study. The participants will be informed that they can ask questions or express their concerns about the study throughout its entire duration and can withdraw at any point. The researchers will also search for signs of discomfort or distress among the participants and will address them individually by talking to the participants and letting them know their options for withdrawal. During the discussion, the participants will be informed that they can refuse to answer questions. The information sheets contain the contact information of all of the members of the research team. The participants will be informed that they are free to contact the researchers with questions and concerns even after the study has ended.

Inbuilt support will be embedded in liaison with key members of the clinical team for recruitment, consent and support mechanisms for the participants if the interviews give rise to issues that need to be followed up (see Appendix 2). The researchers and the members of the clinical team will have ongoing communication about recruitment and data collection. The

members of the clinical team will play an instrumental role during the screening and selection process with the purpose of identifying potential participants and guaranteeing their protection. The key worker will be in charge of following up with the participant after the interview to see if any issues need to be discussed and provide appropriate guidance and assistance. This is important as the research may lead specific patients to face and discuss issues that had previously not been raised.

Considerable researcher burden is also possible. Having two researchers will ensure mutual support. There will also be extensive support from the research team, where expertise in methods and cancer care will be closely matched with researcher need and support. Monthly clinical supervision has been costed into our study.

6. User involvement

This aspect presents specific challenges and has been approached as follows: young adults and their families will be involved in data collection, feedback and analysis as an intrinsic aspect of our study; the NCRN Consumer Liaison Group has been approached to determine specific interest amongst members; the proposal will be read and commented on by family members of two young adults who died of cancer; input into patient information sheets and other patient information will be sought from the Young Peoples Reference Group associated with BRIGHTLIGHT (approximately 20 members - young people diagnosed with cancer between the ages of 14 and 25 (current age 18-29), some of whom have worked with us for over three years on various studies); additional input into patient and subject information sheets will be sought from an additional PPI group, the Cancer Partnership Research Group of the Surrey, Sussex and West Hampshire Clinical Research Network; presentations will be made from January 2013 to ‘Kayleigh’s workshop – terminally talkative’ at the annual young people’s conference, Find Your Sense of Tumour. This was first held in 2011 for those receiving end of life. Facilitated by two psychologists, it is now a regular event at the conference. The attendees from 2011 have also set up their own site on ‘Facebook’, moderated by psychologists, and which we can access if more immediate consultation on the study is needed. Through contact with the NCRN Consumer Liaison Group we have identified patients and patient representatives who will comment on the evolving study and contribute to workshops and panels.

We expect there will be very hard to reach participants consequent on limited awareness of End of Life, high levels of family and professional protection or exaggerated denial. Close relationships between researchers and key workers will be developed to improve chances of access. This will be enhanced by the clinical credibility and national profile of the research team.

7. Data sharing plan

BRIGHTLIGHT is approved by the National Information Governance Board reference ECC 8-05(d)/2011. Electronic data from the study will be stored on an NHS server supported by University College London Hospitals NHS Foundation Trust (UCLH). Paper documents will be stored in a locked filing cabinet in the cancer trials research facility at UCLH. Access will be limited to research associates and chief investigator. All digital recordings will be deleted once a written transcript has been produced. The interview and workshop transcripts will be anonymised and password protected. These will be archived, and available for further analysis after publication of the findings of BRIGHTLIGHT on End of Life.

8. Management of the study

The study is sponsored by UCL.
The core project team led by the CI work in close proximity and will meet regularly.
An advisory group will be established to regularly review and advise on study progress.

Timescales – total period 24 months

Month 1-4	Researchers start employment Detailed review of the literature and analysis of National Cancer Data Repository and National End of Life Care Intelligence Network Detailed protocol development
Month 5-17	Workshops and interviews
Month 18-21	Analysis
Month 21	Expert Panel
Month 22-24	Circulation of account to participants, policy makers, commissioners and other stakeholders. Completion of final account, dissemination through local meetings and writing of peer reviewed publications.

References

[1] Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M. Developing and evaluating complex interventions: the new Medical Research Council guidance. BMJ (Clinical research ed. 2008;337:a1655.

[2] MRC. Complex Interventions Guidance. 2008 [cited March 28th 2012]; Available from: (http://www.mrc.ac.uk/Utilities/Documentrecord/index.htm?d=MRC004871)

[3] National Institute of Health and Clinical Excellence. Improving Outcomes in Children and Young People with Cancer. 2005.

[4] Geiger AM, Castellino SM. Delineating the age ranges used to define adolescents and young adults. J Clin Oncol. 2011 Jun 1;29(16):e492-3.

[5] Helen and Douglas House. [cited 28th March 2012]; Available from: www.helenanddouglas.org.uk

[6] Department of Health. End of life care strategy. Promoting high quality care for all adults at the end of life. 2008.

[7] Office for National Statistics. Mortality Statisitics. 2010 [cited 28th March 2012]; Available from: http://www.ons.gov.uk/ons/publications/re-referencetables. html?edition=tcm%3A77-230730

[8] Barr RD. Adolescents, young adults, and cancer--the international challenge. Cancer. 2011 May 15;117(10 Suppl):2245-9.

[9] Department of Health. Better care: better lives. Improving outcomes and experiences for children, young people and their families living with life-limiting and life-threatening conditions 2008.

[10] Department of Health. End of life care strategy. Promoting high quality care for all adults at the end of life. 2008.

[11] Grinyer A, Thomas C. The importance of place of death in young adults with cancer. Mortality. 2004;9(2):114-31.

[12] Kars MC, Grypdonck MH, de Korte-Verhoef MC, Kamps WA, Meijer-van den Bergh EM, Verkerk MA, et al. Parental experience at the end-of-life in children with cancer: 'preservation' and 'letting go' in relation to loss. Support Care Cancer. 2009 Jan;19(1):27-35.

[13] Grinyer A. Palliative and End of Life Care for Children and Young People: Home, Hospice, Hospital. Oxford: John Wiley and Sons 2012.

[14] Cohen-Gogo S, Marioni G, Laurent S, Gaspar N, Semeraro M, Gabolde M, et al. End of life care in adolescents and young adults with cancer: experience of the adolescent unit of the Institut Gustave Roussy. Eur J Cancer. 2011 Dec;47(18):2735-41.

[15] Baker JN, Hinds PS, Spunt SL, Barfield RC, Allen C, Powell BC, et al. Integration of palliative care practices into the ongoing care of children with cancer: individualized care planning and coordination. Pediatric clinics of North America. 2008 Feb;55(1):223-50, xii.

[16] Christenson K, Lybrand SA, Hubbard CR, Hubble RA, Ahsens L, Black P. Including the perspective of the adolescent in palliative care preferences. J Pediatr Health Care. 2010 Sep-Oct;24(5):286-91.

[17] Pritchard S, Cuvelier G, Harlos M, Barr R. Palliative care in adolescents and young adults with cancer. Cancer. 2011 May 15;117(10 Suppl):2323-8.

[18] Wein S, Pery S, Zer A. Role of palliative care in adolescent and young adult oncology. J Clin Oncol. 2010 Nov 10;28(32):4819-24.

[19] Hinds PS, Oakes LL, Hicks J, Anghelescu DL. End-of-life care for children and adolescents. Seminars in oncology nursing. 2005 Feb;21(1):53-62.

[20] Lundh Hagelin C, Seiger A, Furst CJ. Quality of life in terminal care—with special reference to age, gender and marital status. Support Care Cancer. 2006 Apr;14(4):320-8.

[21] Parr JD, Zhang B, Nilsson ME, Wright A, Balboni T, Duthie E, et al. The influence of age on the likelihood of receiving end-of-life care consistent with patient treatment preferences. Journal of palliative medicine. 2010 Jun;13(6):719-26.

[22] Trevino KM, Maciejewski PK, Fasciano K, Greer JA, Partridge A, Kacel EL, et al. Coping and psychological distress in young adults with advanced cancer. Journal of Supportive

Oncology. 2011;http://www.supportiveoncology.net/single-view/aipcoping-and-psychological-distress-in-young-adults-with-advancedcancer/8ea0588008.html.
[23] Hollis R, Morgan S. The adolescent with cancer--at the edge of no-man's land. The lancet oncology. 2001 Jan;2(1):43-8.
[24] Whelan J. Where should teenagers with cancer be treated? Eur J Cancer. 2003 Dec;39(18):2573-8.
[25] National Cancer Research Network.
[26] Pawson R, Tilley N. Realistic Evaluation. London: Sage 1997.
[27] Charmaz K. Grounded Theory: objectivist and constructivist methods In: Denzin N, Lincoln Y, eds. The Handbook of Qualitative Research. 2nd ed. Thousand Oaks: Sage 2000:509-36.
[28] Oreszcyzn S, Carr S. Improving the link between policy research and practice: using a scenario workshop as a qualitative research toll in the case of genetically modified crops. Qualitative Research. 2008;8:473-97.
[29] Hinds PS, Burghen EA, Pritchard M. Conducting End-of-Life Studies in Pediatric Oncology. WJNR 29(4):448-465; 2007.
[30] Walshe CE, Caress AL, Chew-Graham C, Todd C. Case Studies: A Research Strategy Appropriate for Palliative Care? Palliative Medicine 18(8):677-684; 2004.

Appendices

Appendix 1: Phases of research
Appendix 2: Recruitment and data collection
Appendix 3: BRIGHTLIGHT study synopsis
Appendix 4: Sample 1 recruitment
Appendix 5: Sampling
Appendix 6: Review of medical notes
Appendix 7: Hypothetical scenarios

When Cure Is Not Likely - Methodology Protocol

Interviews and Workshops

[REC Ref: 13/LO/1098]

Rationale

This mini protocol outlines the approach to be implemented for the data collection phase (interviews, workshops and analysis). This will be informed by a realist approach which aims to understand the causal mechanisms which generate outcomes, consideration of context and a desire to improve practice and service delivery. Its iterative approach assists the capture of multidimensional aspects of the evaluation of End of Life Care and explores links between context, mechanism and outcome. It will increase our understanding of 'what works, for whom, in what circumstances, in what respects and why?'

WCINL: Aims and Objectives

Methods

Interviews

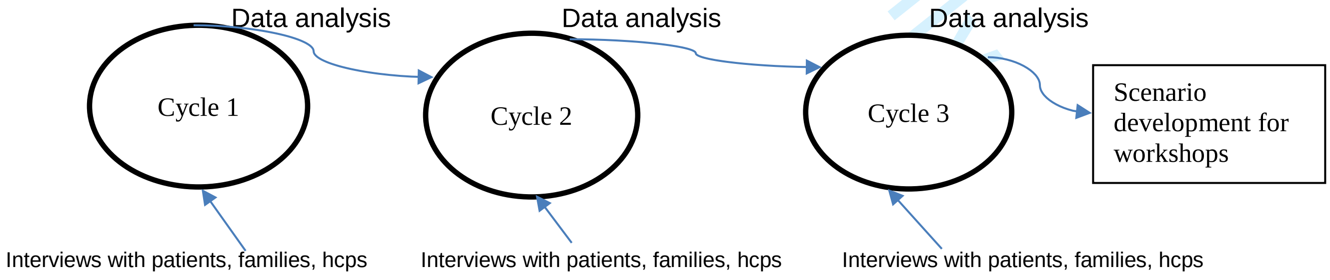
Realistic evaluation will be used as a guiding framework in exploring participants' experiences on the following outcomes;

Good patient centered care
Good family centered care
Responsive clinical care

Our understanding of what is implied by the term 'good care' will develop as our interviews take place and are analysed. For example, emotional spiritual physical; communication, information giving, management of symptoms; promotion of health family systems; social situation; quality of life; supporting finding meaning finding balance between acceptance and hope.

We plan a set of iterative cycles of interviews conducted with 3 groups in each cycle: patients, nominated family members and nominated health care professionals. We shall sample a maximum of 5 patients within each of the two age ranges (16-25 and 25-40) in each cycle, thus a maximum of 15 interviews per age group. Analyses of data will occur after each cycle and findings will be used to inform the conduct of the next cycle of interviews. Once data saturation is reached, the findings will be used to develop scenarios for use in the workshops in the next phase of our work.

Schema of iterative approach for interviews at Time 1:



The interviews will be explorative in their approach due to a significant lack of available evidence about the experiences of this population when cure is not likely and death approaches. This includes a lack of evidence on the availability and appropriateness of palliative approaches to care either alongside treatment or in the dying phase. Through the interviews we aim to explore and understand past and present experiences leading to how the future is viewed and conceptualized. We shall consider the internal, external and reflexive processes of the individual's experience. This will involve the different levels that impact on care: individual,

group (family and within healthcare professional teams), systemic and organizational (Ferlie and Shortell, 2001). We shall also explore the situational and contextual attributes for the individual.

The interviews will be iterative in their style using key questions and prompts to explore the perspectives and experiences of the participant. It is anticipated that each interview will last c. 1 hour. Interview schedules can be found at the end of this document, but in short;

> **Patients** will be asked to discuss their care, and perspectives of support for them, using the broad framework "What has happened, what is happening now, and what do you think will happen next?" Experiences of Internal processes; interpersonal and communication, relationship to others and the situation and context will be explored.
>
> **Family members** (nominated by patients) will be asked about the patient's experiences as well as their experiences and perspectives of support for themselves as a family and/those in a caregiving role. This will include considering the effects on family dynamics, communication between family/ young person and professionals; how families can be supported to enhance their capacity to manage patient care and their perspectives of the past, present and future needs of the patient. Family centered outcomes in loss include experiences in bereavement [could use bereavement risk assessment tools for guidance (Agnew et al., 2010)].
>
> **HCPs** will be asked about their experiences of providing care for the specific patient who nominated them, their more general experiences of caring for patients in this age group when cure is no longer likely leading to discussions to inform our understanding of their internal processes of dealing/coping with younger patients who face death. Here the realist approach will allow both interviewer and interviewee to contribute to discussions of working in an area where sensitive issues are commonplace, thus facilitating opportunities for greater insights.

Patient Age Range: 16-25 years
Cohort 1: BRIGHTLIGHT
Sample size: 15 patients over 3 iterative cycles (maximum 5 patients per cycle)
Sampled from 4 cancer groups – minimum of 3 patients from each group

- leukaemia, lymphoma
- bone and soft tissue sarcoma
- brain and CNS tumours
- carcinomas including germ cell tumours

Interviews in this cohort will take place at time 1, and again after 2-4 months (time 2). The second interviews will explore the current situation and what has changed for the patient since the first interview using the same interview schedule but with a greater focus on what has changed. We anticipate that patients will have experienced both external and internal changes. We shall explore how they have experienced changes in their care, their relationships, and how they now conceptualise the future.

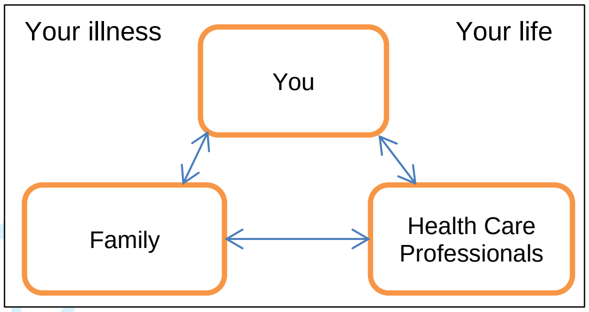
Patient age ranges: 10 16-24 yr olds; 20 25-40 yr olds
Cohort 2: Sites
Sample size: 30: interviews will occur at time 1 only.
Four cancer groups:

- Any carcinomas including breast, colorectal, melanoma, gynaecological and rare tumours – the most frequent diagnoses, so sample to recruit minimum of 21 patients
- Bone and soft tissue sarcoma – minimum 3
- Brain and CNS tumours – minimum 3
- Leukaemia, lymphoma – minimum 3

Conduct of semi-structured realist interviews:
A realist interview allows both interviewer and interviewee to contribute to discussions to facilitate deeper exploration of pertinent issues and allow the possibility of considering innovative issues and solutions.

Interviewers will work to a simple topic guide, allowing free discussion of issues in the past and present and what may occur in the future.

If a patient has difficultly answering questions or conveying their experiences a simple visual prompt may be used (example below) to provide a focus



Simple visual interview prompt

Analysis
All interviews will be audio-taped and transcribed verbatim. Two members of the research team will read in depth each transcript.

A modified grounded theory approach to analysis will be used (Charmaz, 2006). This will take an interpretative approach to identify themes and look for meanings and relationships within the interview data.

Each patient (cohort 2) will generate a set of 3 interviews which will be considered together as case studies which will then be compared and contrasted. In addition, the full set of each category of interviews (patient, family and HCP) will be analysed separately to extract common themes. We shall compare the datasets for the two age groups and look for commonalities and differences. Our findings will be used to inform the development of scenarios to be used in the workshops in the next phase of our research.

We shall also use our findings to develop further our programme theories. These theories have begun with the underlying hypotheses and enablers and barriers that we have identified at the start of our work from the literature and expert opinion of the research team. By defining our proposed outcomes as the provision of 'good' experiences of care for patients and families, and responsive care from HCPs, we shall use our data to develop a fuller understanding of what constitutes good and responsive care, and good experiences for patients and families. We shall use diagrams, flow charts, maps and memos to develop a full picture.

Outline of next stage of research:
Scenario workshops
Workshops will be held across our three recruitment sites across UK, Leeds, London and Southampton. Three workshops will be held at each site (total of 9) comprising of 1 with family members; 1 with healthcare professionals and 1 mixed family and HCP. The scenarios will be used to facilitate interactive discussion of issues arising for patients, families and HCPs in the experiences of care and illness for people in the age groups 16-25 and 25-40 years. The realist approach allows the opportunity for participant to disclose perspectives, opinions and experiences. The conduct of the workshops will enable the generation of hypotheses on mechanisms by which care in the last year of life for people aged 16-40 years might be delivered to enable 'good' care. We shall use our programme theories to underpin our thinking in the workshops and in the analysis of the data they generate. We shall use the emerging hypotheses to populate in more depth our existing programme theories. The teacher-learner approach by which researchers and participants both contribute to discussions will enable the researchers to use their theories and knowledge from the interviews to interact with workshop participants as well as confirm or falsify theories.

References

Agnew, A., Manktelow, R., Taylor, B. and Jones, L. (2009) Bereavement needs assessment in specialist palliative care: a review of the literature. *Palliative Medicine*, vol. 24:1, pp. 46-59.

Charmaz, K. (2006) *Constructing Grounded Theory: A Practical Guide Through Qualitative Analysis*. Sage, London.

Ferlie E.B, Shortell S.M. (2001) Improving the quality of health care in the United Kingdom and the United States: a framework for change. *The Millbank Quarterly*, vol. 79:2, pp. 281-315.

When Cure Is Not Likely – Interview Topic Guides

We need to collect some background information about each participant, so we can describe who has taken part in the study.
What year were you born?
How would you describe your gender?
Do you recall when you were first diagnosed (date/month/year?)
At the moment are you employed (on sick leave) or in full time education?
How would you describe your ethnicity?

During the interview, we would like to talk about a number of issues relating to your medical care, how this may have affected your social life, how well you think information has been given to you or how this could be improved and how decisions have been made about your care;

BOLD = questions Plain text = prompts

Patient	Past (up to WCINL) **Can you tell me what has happened up to this point?**	Present (WCINL) **What is currently happening?**	Future (Post Interview) **Do you think about things that might happen in the future?**
Medical Management	When did you find out something serious was wrong? [diagnosis] When did this happen, where, who was there, how did you feel What treatment did you have [if prompted by patient] How did you find out/ know things were not going so well [prognosis]	How do you think things are going with your illness /symptoms/ cancer at the moment? **Are you receiving any treatments now? Are you on medications, if so what are they for?** **What/who is helpful about the care you are receiving? What/who is unhelpful??** **How do you feel in yourself now? [feelings about medical aspects/situation]**	If yes, what do you think about / how do you plan?? Has anything been discussed with you about potential future treatment plans/ options [this needs to be dependent on what they say about the present]
Social Management	**How did you tell others (family/friends/colleagues) what was happening and how did they react** (illness, treatment, prognosis) **How did you find telling other people about your situation? If it was difficult, which parts were difficult??**	**Thinking about your day to day life – can you do the things that you want to do? If not, what is the impact on your life** **Does your illness have effects on you financially**?	**Is there anything in particular you want to do or achieve? (day to day; immediate, longer term)** **Do you have any worries/fears**

	On a very practical level, what were the biggest things that changed with the illness – e.g. where you were living / work / finance etc. **Have you used the internet to find out information about your illness** (What has been helpful/unhelpful? (illness, treatment) **Do you use social media like Facebook and Twitter? How has your illness affected that??**	**How does your current health affect you your relationships? (sexuality/family/friends/others/dependence/emotional)** **If you have a bad day, who/where do you turn to for support (what do they give you/how do they help? Have you found anything/ anyone particularly supportive)** **How do you feel in yourself now? [emotional wellbeing]** **Have you found a way to make some sense of what's happening at the moment? What has been helpful/unhelpful (spiritual)**	What are your fears (do you worry about) for the future? (parents/children/siblings, increase symptoms, being less able to do things you want to do, decreasing independence, dying process, life after death) **Do you have any hopes, dreams aspirations?**
Communication/information giving	**What was helpful / unhelpful about the conversations you have had with health professionals? How could it have been done better??** (e.g. timings / communication / information)	**Can you tell me about the support you are currently receiving from health care professionals (Cancer CNS, Pall Care, Consultant, GP, District Nurse, Social Worker)** **Can you tell me about the support you are receiving from your family (emotional, practical, financial, care)**	**What have your clinical team said about your future?** **What have you asked your clinical team about your future?** **Who do you think you would contact if you if you have concerns about the future? Why that person?**
Decision making	**Who has been involved in the decisions about your illness / care**?	**Are there any key decisions you are facing at the moment?**	**Are there any key things you think you will need to make decisions about in**

	What input have you had in decisions made about you illness/care?	**(further/stopping treatment; ACP; breaking news to others e.g. small children)** Prompt: If so, is anyone helping you with this? Is there anyone you would like to help you? Are you able to talk to that person / people… if not why not	**the coming weeks and months?** Who do you think might be able to help you with this? Are you able to talk to those people? – if not why not, **Are there questions you want to ask but don't feel able to? What might help to make that easier??** Prompts: (Simple everyday things/activities; Preferred place of care; Preferred place of death; memory boxes, photos, Writing a will; Funeral planning)

Is there anything we haven't mentioned that you would like to talk about today?

Family

We need to collect some background information about each participant, so we can describe who has taken part in the study.

How old are you?
How would you describe your gender?
Do you recall when you were first diagnosed (date/month/year?
At the moment are you employed or in full time education?
How would you describe your ethnicity?

During the interview, we would like to talk about a number of issues relating to x's medical care, how this may have affected their and your social lives and interactions, how well you think information has been given to x or to you and whether this could be improved and how decisions have been made about x's care.

Family	Past **What has happened up to this point**	Present **What is currently happening**	Future **What do you expect to happen in the future…**
Medical Management	When did you find out something serious was wrong with x [diagnosis] When did this happen, where, who was there, how did you feel **How involved have you felt in their treatment and care?** **How would you rate x's experiences of the care (diagnosis, treatment,;** helpful **/ unhelpful –in terms of care delivery / care pathway?** **Were you/other family members offered any support at this time by the clinical team (explain illness, treatment)** What was offered, did this differ by family members what was helpful/unhelpful	What are your views towards the current care that x is receiving How do you feel x is coping at the moment **How do you feel at the moment?** Do you feel supported Is there anyone who is supporting you? Do you have anywhere to go or anyone to talk to if you feel low? **How are the rest of your family coping (explore individual members)** **Is there anything more or different that could be done to help or support you or other family members?**	**Has anyone offered support to think about the future? If yes – was it helpful / unhelpful? If no – would you want some support ? who from? What should it look like?? What would you like?** What do you think might be available to help you and the patient in the future? (Pall Care, CNS, GP) What worries or hopes do you have about this?

	[if prompted by family member] How did you find out/ know things were not going so well [prognosis]		
Social Management	**What was the impact of x's illness on you/your family's day to day life (changes in relationships; priorities; carry on as 'normal')** In what ways has x's illness changed your family (closer, distant) **Who did you tell that x had cancer; how did they react; how did this make you feel?**	**What is the impact of x's illness on day to day life (financial, emotional)** How do you think this has changed your relationship with a) x b) other family members Have you found a way to make some sense of what's happening at the moment? How has this been? **Is there anything particular that sustains you when things are not going so well? (spiritual)**	**What are your fears/ hopes (do you worry about) for the future?** What support do you think you will need in the future (emotional, financial, care) What support do you think other family members may need
Communication/information giving	**What information were you given after x's diagnosis; who gave you this information; what did they say; what this information sufficient?** Do you think they were the best person to tell you this, if not, who would have been better How was information communicated between a) you and x; and b) between the different members of the family What was helpful/ challenging **Were there any things that you**	**What do you know about x's current situation** **What else would you like to know about their current situation; where/who would you go to find this out?** **Who do you turn to for support? Is it adequate, how could it be improved?? What should change??** Who do you share how you are feeling with (family, friends, counsellor); what do they provide	**Has x or their clinical team discussed with you about what might happen in the next weeks and months?** **Have you been offered any support for now or in the future? Is it adequate, how could it be improved? What should change?**

	couldn't talk about to some people; any people you could talk openly too?		
Decision making	**How involved were you in any decisions that x had to make about their treatment** Who decided on your level of involvement Would you have liked greater or less involvement?	**How involved are you in decisions that x has to think about or make (what are these decisions; what involvement have you had)**	**Have you thought about what will happen in the future?** Have you discussed the future with x; what have you talked about (Preferred place of care; Preferred place of death; memory boxes, photos, Writing a will; Funeral planning)

Is there anything we haven't mentioned that you would like to talk about today?

Heath Care Professional

We need to collect some information about each participant.
How old are you?
How would you describe your gender?
Could you tell me what your current job title is please?
How long have you been in this position?
How long have you worked in palliative care?
How long have you worked with the young adult population?
Have you completed any further training for working with young adults with cancer?

During the interview, we would like to discuss the past, present and future dimensions of care for x, and then talk about your wider experience of working with this patient population. With these topics in mind;

With reference to the patient:

- **Past:** How long have you known the patient; What has happened up to this point? (Diagnosis, treatment)
- **Present:** What does the patient/ family understand about what is happening; at what points has communication of significant issues around what is happening taken place, can you tell me more about these. What discussions about this patient have taken place across the MDT
- **Future:** What do you expect to happen in the future? (Prognosis, towards end of life); what has been put in place for the patient – ACP? What support do you think the family will need – has anything been put in place? What is difficult for the patient?
- What do you think would improve the pathway of care for this patient?? How should/could it be done differently?

Wider experiences of working with young adult population

- What are the most important aspects of care for this patient group? (holistic; pain/symptoms; family issues; socio-demographic; financial, Peers, life tasks, support, advocacy, listening, empathy compassion , being there)
- Beyond providing medical care, what other needs do this patient group often have and how do you identify and address these e.g. social, family, educational, financial needs do you think that your system is robust enough, or is there the potential for issues areas to be missed / forgotten?
- At what stage do you know ‘when cure is not likely’; can you give me examples of breaking this news to a patient/their family - where it went well and an example of where it was more challenging. What can make these scenarios more difficult
- How do palliative treatments e.g. chemotherapy affect patients’ desired end of life outcomes (Ref, USA evidence chemo in last months of life associated with CPR ventilation and dying in ICU, Wright et al, 2014)
- How do you negotiate patient/family relationships (e.g. family dynamics)
- What do you enjoy about working with this population?
- What do you find more difficult about working with this population

- How does it affect you when a relatively young cancer patient dies (emotional impact; coping mechanisms; specific patients e.g. those with young children)

- Have any personal or professional experiences affected your practice (e.g. using experiences to improve care; managing emotions; maintain a professional distance)

- Do you have clinical supervision/support available; do you use this or other sources of support? How else do you relax and gain perspective

- Any issues you are aware of generally for professionals, teams, units, places of care, caring for young adults with cancer who may die

- What could be put in place to improve end of life care for this specific patient group?

Are there any other aspects of care for this population that we have not discussed and you feel are important?

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Understanding care when cure is not likely for young adults who face cancer: a realist analysis of data from patients, families and healthcare professionals.

Supplementary file 3 - Our approach to a realist logic of analysis.

Data analysis involved the use of a realist logic analysis with the goal of using the collected data (e.g. interviews) to confirm, refute or refine (test) aspects of our preliminary programme theory. Analysis required interpretation and judgement of data. Data coding was to be deductive (informed by our preliminary programme theory), inductive (arising from the data within data sources) and retroductive (where inferences are made based on interpretations of the data within data sources about underlying causal processes – i.e. mechanisms). We used a series of questions to help us analyse the data, as set out below:

Relevance:
- Are sections of text within the collected data that are relevant to programme theory development or testing?

Interpretation of meaning:
- If the section of text is relevant, do its contents provide data that may be interpreted as functioning as context, mechanism or outcome?

Interpretations and judgements about Context-Mechanism-Outcome-Configurations:
- For the data that has been interpreted as functioning as context, mechanism or outcome, which Context-Mechanism-Outcome-Configuration (CMOC) (partial or complete) does it belong to?
- Are there further data to inform this particular CMOCs contained within this source or other sources? If so, which other sources?
- How does this particular CMOC relate to other CMOCs that have already been developed?

Interpretations and judgements about programme theory:
- How does this particular (full or partial) CMOC relate to the programme theory?
- Within this same source are there data which informs how the CMOC relates to the programme theory? If not, are there data in other sources? Which ones?
- In light of this particular CMOC and any supporting data, does the programme theory need to be changed?

Data to inform our interpretation of the relationships between contexts, mechanisms and outcomes were sought not just within the same data source, but across sources (e.g. mechanisms inferred from one source could help explain the way contexts influenced outcomes in a different source). Synthesising data from different sources is often necessary to compile CMOCs, since not all parts of the configurations will always be articulated in the same source.

Within the analytic process set out above, we used interpretive cross-case comparison to understand and explain how and why observed outcomes have occurred, for example, by comparing contexts where young adults had a 'better' end-of-life care experience with those where this was not to case. This enabled us to understand how context had influenced outcomes and why. When working through the questions set out, where appropriate we used the following forms of reasoning to make sense of the data:

- Juxtaposition of data: for example, where data about context in one source enabled insights into data about outcomes in another source.
- Reconciling of data: where data differed in apparently similar circumstances, further investigation was appropriate in order to find explanations for why these differences had occurred.
- Adjudication of data: on the basis of the plausibility of what was reported.
- Consolidation of data: where outcomes differed in particular contexts, explanations were constructed of how and why these outcomes occur differently.

During the evaluation, we moved iteratively between the analysis of particular examples, refinement of programme theory, and further data collection to test particular theories.

Reporting checklist for qualitative study.

Based on the SRQR guidelines.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the SRQR reporting guidelines, and cite them as:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. Standards for reporting qualitative research: a synthesis of recommendations. Acad Med. 2014;89(9):1245-1251.

		Reporting Item	Page Number
	#1	Concise description of the nature and topic of the study identifying the study as qualitative or indicating the approach (e.g. ethnography, grounded theory) or data collection methods (e.g. interview, focus group) is recommended	6-7
	#2	Summary of the key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results and conclusions	4
Problem formulation	#3	Description and signifcance of the problem / phenomenon studied: review of relevant theory and empirical work; problem statement	6-7
Purpose or research question	#4	Purpose of the study and specific objectives or questions	6-7
Qualitative approach and research paradigm	#5	Qualitative approach (e.g. ethnography, grounded theory, case study, phenomenolgy, narrative research) and guiding theory if	6-7

		appropriate; identifying the research paradigm (e.g. postpositivist, constructivist / interpretivist) is also recommended; rationale. The rationale should briefly discuss the justification for choosing that theory, approach, method or technique rather than other options available; the assumptions and limitations implicit in those choices and how those choices influence study conclusions and transferability. As appropriate the rationale for several items might be discussed together.	
Researcher characteristics and reflexivity	#6	Researchers' characteristics that may influence the research, including personal attributes, qualifications / experience, relationship with participants, assumptions and / or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results and / or transferability	n/a – semi-structured interviews and workshops
Context	#7	Setting / site and salient contextual factors; rationale	7
Sampling strategy	#8	How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g. sampling saturation); rationale	7-8
Ethical issues pertaining to human subjects	#9	Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	10
Data collection methods	#10	Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources / methods, and modification of procedures in response to evolving study findings; rationale	7-10
Data collection	#11	Description of instruments (e.g. interview guides,	7-10,

instruments and technologies		questionnaires) and devices (e.g. audio recorders) used for data collection; if / how the instruments(s) changed over the course of the study	supplementary upload
Units of study	#12	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	10
Data processing	#13	Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymisation / deidentification of excerpts	9
Data analysis	#14	Process by which inferences, themes, etc. were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale	9
Techniques to enhance trustworthiness	#15	Techniques to enhance trustworthiness and credibility of data analysis (e.g. member checking, audit trail, triangulation); rationale	9-10
Syntheses and interpretation	#16	Main findings (e.g. interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	12-16
Links to empirical data	#17	Evidence (e.g. quotes, field notes, text excerpts, photographs) to substantiate analytic findings	12-16
Intergration with prior work, implications, transferability and contribution(s) to the field	#18	Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application / generalizability; identification of unique contributions(s) to scholarship in a discipline or field	21-25
Limitations	#19	Trustworthiness and limitations of findings	25

Conflicts of interest	#20	Potential sources of influence of perceived influence on study conduct and conclusions; how these were managed	25
Funding	#21	Sources of funding and other support; role of funders in data collection, interpretation and reporting	Cover sheet and upload

Understanding care when cure is not likely for young adults who face cancer: a realist analysis of data from patients, families and healthcare professionals.

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2

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Author Contributions

CK, NN, FG, MF, LJ, SP, GW, KMB, SH, RH, AH, LCS, RMT, AT, JW were all involved in study design, review of results and review of this manuscript. Interviews and analysis were conducted by CK and NN: aided by data analysis meetings with LJ, SP, FG, and GW. Workshops were attended by CK, NN, MF, LCS, RH, FG, JW: KB and ABH attended 1 workshop and led another. Participant recruitment was assisted by MF, KB, SH, AH, LCS, JW.

Data sharing statement

No data from interviews with patients, families and health care professionals is available in the public domain given the potential for identification of sensitive information.

Abstract

Objectives: To understand the experiences of young adults with cancer for whom cure is not likely, in particular what may be specific for people aged 16-40 years and how this might affect care.

Design: We used data from multiple sources (semi-structured interviews with people with cancer, nominated family members and healthcare professionals, and workshops) informed by a preliminary programme theory: realist analysis of data within these themes enabled revision of our theory. A realist logic of analysis explored contexts and mechanisms affecting outcomes of care.

Setting: Three cancer centres and associated palliative care services across England.

Participants: We aimed for a purposive sample of 45 people with cancer from two groups: those aged 16-24 years for whom there may be specialist cancer centres and those 16-40 years cared for through general adult services; each could nominate for interview one family member and one healthcare professional. We interviewed three people aged 16-24 years and 30 people 25-40 years diagnosed with cancer (carcinomas; blood cancers; sarcoma; central nervous system tumours) with a clinician-estimated prognosis of less than 12 months along with nominated family carers and healthcare professionals. Nineteen bereaved family members and 47 healthcare professionals participated in workshops.

Results: Data were available from 69 interviews (33 people with cancer, 14 family carers, 22 healthcare professionals) and six workshops. Qualitative analysis revealed seven key themes: loss of control; maintenance of normal life; continuity of care; support for professionals; support for families; importance of language chosen by professionals; financial concerns.

Conclusions: Current care towards end of life for young adults with cancer and their families does not meet needs and expectations. We identified challenges specific to those aged 16-40 years. The burden that care delivery imposes on healthcare professionals must be recognised. These findings can inform recommendations for measures to be incorporated into services.

Strengths and limitations of this study

1. In response to the lack of empirical research, policy and expert practice to inform delivery of optimal care for young adults when cure of their cancer is not likely, we collected data directly from patients with incurable cancer, and their nominated family carers and healthcare professionals..
2. We used realist evaluation to seek the underlying mechanisms in our data and how these influenced outcomes.
3. People with blood cancers and those aged between 16-24 years were difficult to recruit and may have unrecognised specific needs.
4. Although analysis of this unique data set has highlighted specific challenges for young adults, their families and healthcare professionals in the delivery of end-of-life care, additional work is needed to make changes to practice that will improve experience and outcomes.

Background

Cancer in young adults under 40 years is notable because it comprises a wide range of malignancies, has specific challenges to improving both length and quality of life, but is relatively uncommon.(1) One quarter of all deaths in the United Kingdom in people aged 16-40 years are from cancer.(2) In Europe there are over 27,000 deaths per year in this age group.(3) Despite increasing empirical evidence of the specific needs of young adults in specialist cancer care, there is little evidence about their experiences towards the end-of-life. (4-6)

Studies of adults with cancer usually cover a wide age range with most participants aged over 40 years. The existing literature tends to summarise good practice and, where studies have been undertaken, little evidence comes directly from people with cancer. (7-10) Given the identified gap in current literature, this research aims to contribute to Ngwenya et al.'s conclusion that "Future research should focus on age-specific evidence about the end-of-life experiences and preferences for young adults with cancer and their informal carers". (5)

Concerns about improving end-of-life care are not confined to young adults. A recent interdisciplinary report published by the Royal College of Physicians in the UK summarises the concerns expressed by professionals, patients, families and other stakeholders such as charities. This report suggests that much more can be done to overcome barriers and myths that have been long-identified. The value of the perspective brought by patients and families is highlighted as a means to bring timeliness and honesty to discussions about dying whilst at the same time accounting for and respecting specific circumstances set by factors such as underlying disease, faith and as addressed here, age. (11)

Boundaries between curative and palliative cancer treatments are often blurred as decisions may be influenced by cancer type, age and family circumstances as well as the experience and skills of healthcare professionals (HCPs). Avoidance and delaying of discussions about planning for care as heath deteriorates and end-of-life decisions are common, often affecting the quality of care.(12) Professionals consistently acknowledge the challenges of managing end-of-life care for younger people, which may have commonalities with and, importantly, differences from those people with cancer at older ages as death approaches. (9, 10, 13)

This work considers both the problem of limited data available in the literature and the desirability of understanding the experience of facing a poor prognosis at a young age from multiple perspectives. We wished to understand what were the core components in the pathways of care in the last year of life for people with cancer aged 16-40 years; whether there were any differences between the experiences of people with cancer from the age ranges 16-

24 and 25-40 years; how young adults and their families can be supported in the last year of life to achieve their preferences for care; and what challenges exist for health and social care professionals providing care.

To develop our knowledge of end-of-life care in adolescents and young adults aged 16-40 years (referred to in this paper as ‘young adults’) with cancer, we sought to collect data directly from young adults who were facing a poor prognosis, their families and HCPs involved in their care. To gain a deep understanding of the contexts that may be specific to this age group, we chose to explore our data using a realist evaluation approach.(14) A realist evaluation approach focuses on explanations, taking account of contexts and mechanisms that may affect outcomes. It addresses questions about what works for whom, in what circumstances and in what respects, and how?

Consistent with the realist evaluation approach, we began our research with a preliminary programme theory. A programme theory is a description, in words or diagrams, of what is supposed to be done in a policy or programme (theory of action) and how and why that is expected to work (theory of change). (15) Details about how to develop programme theories is beyond the scope of this paper but methodological guidance is available.(16) Our preliminary programme theory was informed by expert opinion within our research team which was led by clinical academic specialists in the care of young adults with cancer. Our thinking was also informed by a narrative review of the existing literature, Phase i of our study, previously reported.(5) A preliminary programme theory provides an initial framework of understanding for the area of research being considered. Being preliminary it is, by definition, subject to iterative change and refinement based on the data we collected and analysed. We anticipated that some elements of our preliminary programme theory may be strengthened and others refuted; indeed, new elements may emerge that require significant additions to what is thought to be our best understanding at the outset. At the end of the project our expectation was that we would be able to develop and confirm, refute or refine aspects of preliminary programme theory and ensure that it is more realist in nature. That is, we wanted to ensure that at the close of the project we had a programme theory that contained as many realist causal explanations (i.e. consisting of embedded Context-Mechanism-Outcome configurations) within it as was possible.

Our preliminary programme theory was:

‘That there are specific differences in experiences of and preferences for care towards the end-of-life for those with cancer aged 16-24 and 25-40 years compared to those who are older. Life-threatening illness in the young is untimely, it disrupts expected biographies, and

maintaining a sense of control and normality in everyday life may be important. The role of close family members is complex and integral to the experiences of the person with cancer.'

We used this theory to develop topics for use in semi-structured interviews with young adults with cancer, family members and HCPs, and to underpin scenarios used in workshop discussions with HCPs and bereaved family members. That is, our preliminary programme theory sets out our initial hypotheses of the differences we thought were likely to set apart the end-of-life care experiences and preferences for younger people. Our interviews were thus developed by the project team in such a way as to be able to gather data that would enable us to confirm, refute or refine aspects of our programme theory. For example, because we hypothesised that a sense of control might influence end-of-life care experiences, we deliberately developed interview questions that asked about this issue. An important point about our initial programme theory is that it was refined as the evaluation progressed based on data gathered. As such, our expectation was that our preliminary programme theory would need to be refined to have adequate explanatory value.

In this paper, we describe data arising from these interviews and workshops. We used our data analysis to further explore and develop realist causal explanations that may explain parts of our preliminary programme theory. As is expected in realist evaluations, as the evaluation progressed, we developed a revised programme theory that can be used to underpin recommendations for policy and practice and inform future research.

Methods

A multi-method realist study was undertaken (Figure 1). A realist evaluation approach was used as we wanted to explain and understand contextual influences on the experiences of and preferences for care towards the end-of-life for those with cancer aged 16-24 and 25-40. Here we report on Phases ii-iv, using RAMESES standards for reporting realist evaluations. (17) Phase v will be reported separately.

Recruitment and participants

We aimed to recruit a purposive sample of young people aged 16-40 with cancer, in two cohorts with an expected prognosis of less than one year, across four cancer groups: carcinomas; leukaemia and lymphoma; bone and soft tissue sarcoma; and central nervous system (CNS) tumours, which account for more than three-quarters of cancers occurring in this age group. Estimation of prognosis was made at each site by clinicians involved in screening and identifying people with cancer for the study. They used clinical records, their own clinical knowledge of disease progression and liaised with other members of the clinical team to confirm, at the time of approach, that the prognosis for each individual was likely to be less than one year. In cohort 1 we planned to recruit a maximum of 15 participants aged 16-24 years, including a minimum of three participants from each of the cancer groups, to be interviewed at two time points; recruitment began via a national cohort study investigating whether specialist cancer services add value (www.brightlightstudy.com) and was later extended, due to poor recruitment, to include five principal treatment centres and a hospice for young adults. Cohort 2 was recruited from three specialist cancer services and three hospices in England and consisted of a maximum sample of 30 participants between the ages of 16-40. All cohort 2 participants were invited to nominate a family member and HCP involved in their care for interview. The first-hand clinical experience of many in the project team aided the development of the study. Knowing that this is an under-researched population within the context of the study and drawing on professional experience to guide data collection, analysis and interpretation was essential. Further details are available in the protocols (supplementary files 1 and 2).

Data collection

Semi-structured interviews

All participants took part in a semi-structured interview at a single time point using a topic guide. Cohort 1 participants were invited to take part in a later second interview.(14) The topic guide was developed from a review of the limited existing literature for the 16-40 age range (5) and the clinical and academic expertise within the project team who work directly with this population. We sought patient and public involvement (PPI) input to refine the topic guide coverage and phrasing of the questions, which explored medical, social, communication and decision-making experiences for people with cancer and their families. We asked HCPs to reflect on the care of the person with cancer and their practice with those approaching the end-of-life.

Workshops

We held workshops in London, Southampton and Leeds. The workshops involved the participants sitting as one group. One clinical member of the team acted as the facilitator for the HCP workshops and two clinical members of the team were co-facilitators for the bereaved relative workshops. The co-facilitation meant that if someone from the group needed to leave or have a break from the discussion they could be supported by one of the co-facilitators whilst the workshop was able to continue. At the start of the workshop the facilitator introduced the study, outlined the workshop and informed consent obtained. The HCP workshops focused around the scenarios and the perspectives of different professional roles. The bereaved relative groups were guided by one of the facilitators with the participants sharing narratives around their experiences with other participants either supporting the narrative or outlining how their experience differed.

1. Healthcare professionals

Three workshops involved HCPs working in both hospital and community settings who were recruited by the participating sites. Two scenarios were developed from initial interview analysis and reported experiences (Table 1). We sought to present contrasting fictional patients differing by age, gender and social situations which had raised a number of common issues arising from the interview data that the workshop participants were asked to discuss.

2. Bereaved relatives

We held three workshops with bereaved relatives who were invited to take part by bereavement services in participating hospices. The use of scenarios for this group were felt to be too abstract; and so these workshops focused on the relatives' individual experiences. The workshops involved open discussions and sought to collect information that had not emerged previously in the interviews, particularly concerning the last days of life.

Table 1. Scenarios used in health care professionals' workshop

Scenario 1: 16-40 year old patient with haematological malignancy	**Scenario 2: 25-40 year old patient with oncological malignancy**
Mannu, 19, diagnosed with Hodgkin's disease in December 2013. Between December and June treated with curative intent. Relapsed June 2014 – no sibling bone marrow donor available – deteriorated before one could be found. Social Science student – sporty. University not local. Friends all at University. School friends all over country also at University. Keeps in touch with friends via Facebook. Home Returned to live with Mum, Dad and sister aged 12. Grandparents supportive – all aware of diagnosis and prognosis. Sikh faith. Supportive in background. Home is a three bed semi with a bathroom upstairs and downstairs toilet. November 2014 Inpatient. Deteriorating – wants to be at home. Unable to do stairs therefore need to make adaptations. • Symptoms – shortness of breath, cough and fatigue. • Care – family keen to do.	Helen, 38, diagnosed with colon cancer in May 2014. Helen lives with her partner and their 18 month old baby. Soon after diagnoses she had surgery for a stoma fitting and was diagnosed with liver metastases a few weeks after. Social She has support from her parents, brother and her partner's parents. She is currently on sick leave and misses friends from the office. They have reduced income due to her being on maternity leave before her diagnosis, although she has critical Illness Policy which will pay off their mortgage and so this is reassuring for her. Home She lives an hour's drive from her parents in a duplex house with stairs. She is getting more symptomatic and experiencing fatigue. Partner is concerned about coping with a young child and partner as she deteriorates. September 2014 Helen has lost weight and is aware that she is getting weaker and has difficulty picking up/carrying her child. She is currently on a

- Discharge home with Community Palliative Care Team input. - Contact with charities - Willow Foundation, CLIC Sargent December 2014 Increased fatigue. Treated with radiotherapy to chest. Cough and fatigue. January 2015 Further deterioration. Bed bound. Home oxygen. Anticipatory medications.	24/7 syringe driver and the District Nurse visits daily. Referral to hospice palliative care has been made but she has not yet been in contact. Helen is referred for a clinical trial as still relatively well and no conventional treatment options. December 2014 Chemotherapy stopped as disease not responding - parents devastated. Parents not able to access psychological support as they live 'out of the area' Advanced care planning with clinical nurse specialist causes tension as parents do not wish Helen to be 'not for resuscitation' February 2015 House requires adaptations due to her physical condition. Increasingly housebound due to steps and steep hill Partner feels he can no longer cope as Helen's condition deteriorates further.

All patient, family and HCP participants were provided with a Participant Information Sheet which outlined the study, their expected involvement and the right to withdraw at any point. Written informed consent was obtained from all those who participated in the study. Interviews and workshops were audio-recorded, transcribed verbatim and anonymised before analysis. Field notes were recorded during the workshops.

Data Analysis

Data were entered into a qualitative analysis software programme, NVivo 10 to facilitate analysis. (18) A realist evaluation approach enabled us to identify and understand (a) the outcomes for young people receiving care; (b) when these outcomes were likely to occur (the contexts); and (c) why (the mechanism).(14) Our analysis was multi-staged (figure 2):

- *Stage One - identification of emergent themes.* Charmaz's grounded theory approach was used.(19) Initial codes (summary of what participants were describing) were open and inductive from the data using verbatim quotes or researcher-generated codes to inform a conceptual framework. We then developed categories by grouping similar codes. The categories were identified by two researchers working independently. Emergent findings were discussed within the wider research team and further refined into themes.
- *Stage Two - realist logic of analysis.* This stage was undertaken as we wanted to develop findings that had a clear warrant for transferability. In other words, by reanalysing our themes, using a realist logic of analysis, we would be able to identify

the commonly occurring mechanisms within this population group that caused the outcome patterns we had found. The way we operationalised a realist logic to develop CMO configurations may be found in supplementary file 3.

Re-analysis and re-interpretation of the themes to develop CMO configurations was undertaken by CK and NN aided by data analysis meetings with LJ, SP, FG, and GW. To assist the re-analysis and re-interpretation process, we attempted to develop CMO configurations that explained the outcomes in as many parts as possible of our preliminary programme theory; of the care pathways and experiences of people with cancer, family members and HCPs. For each of these mini programme theories we re-analysed the data that we drew on to develop each theme to build context-mechanisms-outcome (CMO) configurations - i.e. develop realist causal explanations of outcomes that occurred within different contexts (e.g. social rules and cultural systems). Workshop data were analysed in the same two-step manner and used to confirm, refute or refine the CMO configurations within the 'mini' programme theories.

Ethical review

The study was approved by Central London Research Ethics Committee (Reference: 13/LO/1098) and informed consent was sought from all participants at the time of participation.

Patient and Public involvement

We responded to a funding call from Marie Curie, a UK charitable organisation which provides care and support to people with terminal illnesses and their families, specifically seeking research proposals focussed on the needs of young adults. We sought the views of people with cancer on study design and written information including patient information sheets through the Cancer Partnership Research Group of the Surrey, West Sussex and Hampshire Cancer Network and the National Cancer Research Network Consumer Group. An independent steering committee, which included a bereaved parent of a young adult, provided advice and oversight on study conduct. We plan to work with Marie Curie on patient-focussed dissemination of our findings.

Results

Table 2 summarises the participants by cohort. A total of 69 interviews were conducted (33 people with cancer, 14 family members, 22 HCPs); 19 bereaved family members and 47 HCPs took part across six workshops.

Table 2 Participant Details

Cohort 1 N=30		Cohort 1 N=30	Cohort 2 N=3
Gender	Male	11	3
	Female	19	0
Age	Median (range) years	32 (16-39)	
Ethnicity	White British	19	3
	Any other White background	4	
	Asian/Asian British/Black/African/Caribbean/Black British	7	
Cancer type	Carcinoma	18	1
	Sarcoma	6	2
	Blood cancer	2	
	Other (incl. melanoma/CNS)	4	
Education/Working	Working Part Time	2	
	Working Full Time	2	
	Sick Leave	9	2
	Sick leave from education	2	1
	Not Working/Early retirement	14	
Nominated, interviewed family or other	Husband/Wife/Partner	5	
	Parent/sibling	8	
Nominated, interviewed healthcare professionals	Clinical nurse specialist	13	
	General Practitioner	2	
	Hospital doctor	4	
	Allied Health Professional	3	
	Patient did not nominate	5	
	Healthcare Professional declined participation	3	

The results are presented in three sections:

1. Our thematic analysis of qualitative participant data.
2. Realistic logic of analysis reporting context, mechanisms and outcome (CMO) configurations developed from re-analyses of the themes.
3. The connections and links between contexts, mechanisms and outcomes as leading to the revision of our programme theory.

Section 1 Thematic analysis

Seven key themes emerged each of which is accompanied by one or more illustrative verbatim section of texts from our data.

Loss of Control

As illness progressed and young adults with cancer became more debilitated, they often felt a loss of control over how they lived their lives. This was a shift from independence to a growing dependence on others for physical, emotional, practical or financial support provided by family, friends, HCPs or the wider state. The future became unpredictable and planning was difficult. Maintaining a sense of control and continuing to take part in activities, albeit compromised, was important:

My independence. For me, being able to do things on my own is definitely something that I miss, without - being carefree, I can't be carefree, I can't just go out and have, get drunk with friends any more. I can't go out for a long night and dress up in heels and get bashed about, because I have a port in, I've got cancer, you know, I have to go and sit down at a bar, have a non-alcoholic cocktail. It doesn't mean I can't socialise and have a good time with them, I still do. But I'm uncomfortable when I dress up now, whereas before I had the figure and went to the gym and felt more comfortable in myself. (Cohort 2 – Patient 20)

Maintenance of Normal Life

Participants all desired to continue, as far as possible, living a 'normal life' e.g. working, taking part in activities, looking after their children. Normality provided reassurance and a sense of control but it could also be a defensive response and a shield of denial about the realities of dying from cancer. As the disease progressed the sense of 'what was normal' needed to be reframed and adjusted:

I'm at probably the worst stage I've ever been with this illness, obviously because it's more advanced. Yet people are just saying, "You're looking great." And when I look in the mirror, I don't feel like I've got cancer. I don't feel like – obviously I do because I know that I do, but I don't feel any different to how I used to feel. Obviously yes you've got a few aches and pains and stuff, but you think like, when you hear someone's dying of cancer, you think that person will feel like they are. But like I know that I am, but I don't feel like I am, because it's quite a disconnect of like how – you know, like when you're feeling alright and you're going round doing stuff, and you're just doing stuff like everyone else, you just kind of forget. You go to work and you just have the same sort of, do the same things you were doing before you had cancer. You just forget, I forget sometimes (Cohort 2 – Patient 19)

Continuity of Care

Young adults valued being known by the HCPs involved in their care and preferred a joined-up care pathway between them, the HCPs and other health services. This relied on maintaining continuity of communication and information between HCPs, services and themselves with a shared knowledge of the care plan. They generally preferred to be seen by the same HCPs as they felt they could build rapport and feel known as a person. When they moved between services e.g. from oncology to palliative care or from hospital to hospice, they wanted this to be a joined-up seamless shift:

So we went into this initial meeting and [1st tumour CNS], who is the CNS, was there. And Dr [Consultant] was the one that kept us waiting. And it was said at that point, "[1st tumour CNS] will be your CNS, presumably key worker, throughout this process, she will be at every one of your appointments when you come to clinic." And I was like, great, and he gave me her number and a pack and, you know, I felt quite supported by that. … I understand not being able to the same nurse every time, that's not possible, but like if you had a team that were allocated a certain number of patients – because they just, they don't know you. And I've noticed that across the course of having another lot, you know, and I've really – I've kind of got to know a lot of them because I've been there, you know, over the course of a year. But, you know, it is at the beginning, it's someone different every week. And they don't know anything about you. And I went in expecting them to have read my notes, know what kind of cancer it was, know, you know, some of my background, and totally naively – they – and I think it's unfair to them, they are there just to administer medication (Cohort 2 – Patient 29)

Professionals Need Support

Professionals in either cancer or palliative care settings tended to have greater experience of caring for older adults. They had less experience providing end-of-life care to those aged 16-40 and fewer 'tools' or strategies to offer this younger population. Professionals found caring for young adults as they deteriorated both professionally and emotionally challenging and burdensome, as witness to young people prematurely reaching the end of their lives coupled with a weight of expectation to do more. The availability, accessibility and use of support for HCPs was variable and ranged from peer to professional support with a perception that experienced senior doctors were less likely to be in need. In contrast, nurses were perceived to be more likely to require and/or seek out support:

But there's always been this sort of demarcation that when they come to the – come to, "They're now incurable," they go somewhere else. And that 'somewhere else' is always nebulous. 'Someone else' looks after them 'somewhere else.' Do you know what I mean? … "Oh they go over there now." As I said earlier, the palliative team will look after them. And I *don't think any of us [Oncology CNS] have ever really gone to see what the palliative team do or see how much input they have. And is that a, is that a lack of professionalism or is that a survival mechanism for ourselves? And I have a feeling it's the latter. I have a feeling that it's very much a survival mechanism for ourselves because then we can just close that bit off and we can get on over here. And we'd like to know how they are, but we don't have to be the one that tells them. (Cohort 2 – HCP 16)*

Families Need Support

Families provided multiple types of support (practical, physical, emotional, financial) to the person with cancer to complement or supplement professional care:

Well I do as much for her as I can and I go out and do all her shopping. And if her husband is not around to pick the kids up from school, myself and my other daughter, we sort of take care of the kids. And also we've been taking them out as well because – and that upsets [name] more especially during the school holidays last week that they couldn't go anywhere. And she started saying, "I'm not a good mum." (Cohort 2 – Family 22)

The impending decline and death of a young family member was usually unanticipated and a situation that families have rarely experienced before. Family members generally had fewer appropriate skills to care for the person as their cancer progressed. Families expressed a wish for some form of access to information or training to care for their loved one appropriately. Looking back, bereaved families commented that their skills to deliver care at the end-of-life were limited and they would have liked access to some basic training and emotional support.

Language

The use of language by HCPs to describe an approach to care may not convey the same meaning to young adults with cancer and their families. For example, words such as hospice conjured up particular scenarios and carried ambiguity about the imminence of the end-of-life; such terms were often left unexplained, causing distress:

I do remember him [Consultant] saying, I can't really remember the conversation massively, but I do remember him keep saying, "Tumour, there's a tumour." And then I literally did have to say, "Hang on a minute, do you mean cancer?" and he said, "Yes, we've got to run more tests and this, that and the other, but yes." But that's the only thing I remember really about it, if you know what I mean. (Cohort 2 – Patient 14)

Financial Concerns

There were few participants for whom finance was not a concern. For those who were younger and still in education or training the burden tended to fall on their families. For those who were working, with loans, mortgages or dependents, the impact of cancer compromised their ability to support themselves and their families. Concerns were expressed about changes in lifestyle whereby the basics were prioritised. There was some confusion around entitlement to benefits or equivalent sources of financial support and limited access to tailored financial advice or guidance:

But you could do with somebody saying to you, in the first place, "You need somebody to help you to do this," you know what I mean, you need somebody who can guide you through the system. And I think the same applied with [name]. He'd think, "Oh well I've just got to fill this form in and I've got..." but actually filling those forms in is a damned hard job. (Cohort 2 – Family 23)

You haven't asked to be in that position [dying from cancer]. So I shouldn't have to go to work and think, 'Well I'll do a monotonous job just to pay the bills to only live another few months.' If I've only got a few more months to live, I'd rather spend it with my family, you know, having the time with them. (Cohort 2 – Patient 6)

Section 2 Realist explanations of our themes presented in the form of Context-Mechanisms-Outcome (CMO) configurations

We reanalysed and re-interpreted our emergent themes using a realist logic of analysis. We attempted to identify mechanisms (generative causal processes) that are activated in the contexts we had found within the themes we uncovered. Our interview data were purely qualitative and so likely to be limited in the range of relevant data needed to build CMO configurations. To supplement these data, we deliberately drew on the extensive content expertise of the project team, workshops and where relevant, existing theories on needs of people living with cancer.

Details summarising the CMO configurations are presented in Table 3.

Table 3 CMO configurations, illustrative quotes and summary of our interpretations

Context-Mechanism- Outcome Configuration	Quote	Related theme
CMO 1 The diagnosis of cancer (context), changed the perception of control (mechanism) in young adults to cause distress, frustration and anger (outcomes).	*… like the feeling that I've got control over it, like complete control. For me that's extremely important. As soon as I lose that, I think I'd really struggle. And I need to, yes, feel as though I'm in the driving seat more or less. (Cohort 1 – participant 1)*	From the Loss of Control theme: The unexpected diagnosis of cancer disrupted everyday life and young adults often had to relinquish control and permit others to manage aspects of their life. The feeling of 'loss of control' was experienced throughout the diagnostic and treatment phases and seemed to increase when cure was not likely as participants experienced a loss of their anticipated future.
CMO 2 In the context of disease progression (context), young adults continued with normal activities as a coping strategy that offered distraction (mechanism) leading to a feeling of some kind of 'normality' (outcome). A poor prognosis and physical decline compromised the maintenance of a 'normal life'.	*"Yes but we're not going to do that" he [son] said, "We're just going to carry on as normal." And I thought actually he's right because carrying on normal makes it, it does make it more real. And more memorable … yes we do some lovely things, but it's just trying to keep everything as normal as possible really and just make the most of that time [Cohort 2 – family member – son did not participate]*	From the Maintenance of Normal Life theme: Young adults wanted to live as normal a life for as long as possible. Young adults and their families adjusted to a new normality, to accommodate the changes their disease progression created.
CMO 3 When there was trust between HCPs and a young adult (context), it was easier to introduce change or a new service (outcome) because a sense of	*I think continuity is one thing that I'd put on a pedestal as being the most important, as a patient. It's horrible seeing different people and having to tell your story over and over and over again. Yet when you see somebody you know, and they know*	From the Continuity of Care theme: Young adults reported a high expectation and preference for continuity of care within and between services, which for them meant seeing the same HCPs whenever possible:

abandonment (mechanism) or apprehension (mechanism) was less likely to occur. In contexts where continuity was provided (i.e. seeing the same HCP) trust developed (outcome) because of a sense of being known (mechanism).	*your story, they know whether you're well or you're not well. They know how your psychology works a little bit. So they know how to present things to you. That makes a huge difference to how you trust them, what your relationship is like and how you respond to them [HCP's] (Cohort 2 – Participant 7).*	
CMO 4 When a young person is dying because of cancer (context), HCPs find it challenging to talk about a poor prognosis or a shift in goal of treatment (outcome) because of their personal emotional discomfort of such discussions (mechanism).	*I find some of the younger patients it feels very unfair and I do reflect a lot on my own mortality and how I would cope (Cohort 2 – Participant 1 HCP)* *And it's a very stark contrast that [support] seems to be important for nurses but it's not seen to be important for doctors … as an individual, you don't talk because nobody wants you to talk about it, because you're the strong leader … Some doctors will just completely divorce themselves from it and will not engage in any shape or way with their patients … But I think you just sort of, you potentially just end up with, you know, increasingly tired and burnt out and disengaged doctors (Consultant Oncologist, HCP workshop 1)*	From the Professionals Need Support theme: The loss of a 'life partially lived' can be difficult for professionals, who feel a greater burden of sadness when young adults die HCP workshop participants felt support for them was often reactive rather than proactive with nurses more likely to receive support than doctors. A further distinction was recognised between junior and senior doctors:
CMO 5 When the way services are delivered for young adults does not fully recognise the additional needs of the family or care-givers (context), this leads to	*It is quite upsetting because, we actually felt abandoned, I felt abandoned.* *[Facilitator: By?]*	From the Families Need Support theme: Families often provided informal care for young adults within the home. Whilst they wanted to support their child or partner, caring created a

them feeling marginalised (mechanism) resulting in feelings such as abandonment and distress (outcomes).	*By just the whole system really. It was just, if you didn't ask, you wouldn't know (Family workshop 2- bereaved husband)* *And she [24 yr. old daughter] sort of became more and more sleepy and distant from us. But nobody would say to me, "This is what to look for. When she dies, this is what's going to happen. (Family workshop 3 – bereaved mother)*	further burden. Families felt insufficiently supported in this role.
CMO 6 When emotive language is used in palliative and end-of-life care (context), misunderstandings (mechanisms) can easily occur, leading to a range of different outcomes from encouraging hope through to despair (outcomes).	*So when my breast care nurses referred me to a hospice, I was like "Oh my god, that's horrendous, I don't want to do it". But, you know, it's been one of the best ever things. And I kept putting it off and saying, "I'm not ready for it, I'm not ready for it." And she [CNS] went, "Look, if you just make contact, then when you do need them, you can tap into them and they're quite good at financial advice". So I said, "Okay right let's do it". And actually they've been fantastic. Actually from just the level of sorting things out. (Cohort 2 – participant 19).*	From the Language theme: One example was the use of the term 'hospice'. When this was first raised with young adults their initial reaction was one of rejection as hospices were where older people went to die and young adults did not believe that they were at this stage:
CMO 7 Few young adults have thoughts about long term financial planning as they did not anticipate serious	*You haven't asked to be in that position. So I shouldn't have to go to work and think, 'Well I'll do a monotonous job just to pay the bills to only live*	From Financial Concerns theme: Financial concerns and insecurity are almost always expressed by young adults with cancer.

illness (context). This can lead to individuals and families facing financial precariousness (mechanism). Access to tailored advice, whilst it may not solve financial concerns, may provide individuals and their families a range of 'tools' to better cope with their financial situation (outcome).	*another few months.' If I've only got a few more months to live, I'd rather spend it with my family, you know, having the time with them. (Cohort 2 – Patient 6)* *But, yes, I think that's the crappest thing, it's not having – if you are single and I don't have a rich family, you know, yes it's just the whole worry of like affording things and knowing what kind of life you're going to end up with if you give up work, especially when you've been used to a different kind of life. (Cohort 2 – Patient 19).*	

CMO: context, mechanism and outcome

Section 3 Revision of preliminary programme theory

Our re-analyses of the data enabled us to confirm, further develop and refine aspects of our preliminary programme theory – namely control, normality and family support. We were also able to add to our preliminary programme theory the concepts of continuity, professional support, language and financial support. In what follows, we summarise important aspects of our refined programme theory.

Age specific issues

We now understand that *for those aged 16-40 there are specific differences between the end-of-life care experience and preferences.* However, *rather than being wholly defined by age, the stages in a young person's life course may be a better way to approach, understand and support these differences.*

Maintenance of control and sense of normality

Our data underpin these concepts within our preliminary programme theory. We have learned that *young adults with cancer need support to put strategies in place to retain control and live as normally as possible whist providing a space to discuss and plan for their shortened future.*

Families of younger people with cancer

We found that *the family often are not appropriately equipped to provide the level of care and support that they want to provide during the last year of life of the young adult with cancer and lack the means to be 'skilled-up' for this role.*

Healthcare professionals

We found that *healthcare professionals lack age (16-40-year-old) life course-specific knowledge to develop strategies to support patients in their last year of life and their families.*

Discussion

In this study, we used a realist evaluation approach to gain a deeper understanding of the particular contexts that may be specific to the experiences of young adults aged 16-40 years with cancer as they approached their end-of-life. We re-analysed our initial seven themes into seven CMO configurations that explained the specific needs of the end-of-life experiences of young adults with cancer. The implications of these specific needs are set out below and compared and contrasted with the existing literature.

Life course and not age matters

We found that within this group, end-of-life experiences and preferences cannot be neatly isolated into the two age ranges we studied (16-24 years and 25-40 years). A better way of approaching, understanding and supporting young adults may be to consider where they are in their life course, as there may be more in common, than different, between those with similar life course experiences, for example being in education, maintaining a career, having children or caring responsibilities. The usefulness of taking such an approach is also found in the wider literature on end-of-life care for young adults and so reinforces this finding.(20) Adolescence and young adulthood is a developmental stage when individuals shape their identities, gain autonomy, make career choices and develop intimate relationships. A cancer diagnosis at this stage is “off-time” during the normative life cycle: life is interrupted, developmental tasks and identity formation are challenged and few peers will share their cancer experience.(20) In common with Soanes and Gibson we found that participants across this age range reported a desire to maintain these aspects of their life, as well as their identity for example, as a student, a professional, or parent, in part to maintain a sense of normality and control.(21)

Giving young people the chance to have control and to feel normal

We found, perhaps unsurprisingly, the pivotal role of HCPs in supporting young adults with cancer. However, we were able to identify that an important ‘block’ to the support provided comes from the emotional discomfort felt by HCPs when discussing aspects of care specifically with young adults – such as discussions about prognosis. This is important as a cancer diagnosis creates great uncertainty and the knowledge that there will not be a cure creates a dissonance between the life that was expected and the reality of a life that will be significantly shorter than expected. For emerging adults and early independent adults, as disease progresses, dissonance is also present as their independence is compromised with an increasing and unanticipated dependence on others. This can affect their ability to attend school, college or work as well as taking part in family or social activities or fulfilling caring duties for others e.g. looking after young children. Adaptation is a mechanism through which there is a recognition of what can no longer be achieved due to disease progression.(22) An adapted normality can be achieved together with a sense of control, allowing for realistic goal setting.(23) Advance care planning could facilitate this adaptation. However, few participants in our study reported having had conversations about their options or the care they wanted to receive. Some HCPs avoided such conversations because of the emotional burden to themselves, not wishing to challenge either hope or a young person’s possible denial about their situation. This might be an example of what Bell et al refer to ‘as social constraint’, i.e. words and actions that inhibit end-of-life discussions.(24) A further notable finding from the data indicates that all parties appear to wait for another to raise the topic of end-of-life. The ‘window of opportunity’ (17) often fails to appear, thus in some cases the topic is avoided. This has the potential to delay adaptation and limit the time available for professional support, which could help young adults plan and make as much as possible of remaining time.(25) For those with dependents, particularly young children, delaying adaptation could impact on their roles as parents, delaying the opportunity to prepare and create memories for themselves and their families.(23) When end-of-life was addressed, this tended to be when health had deteriorated, and that window of opportunity, albeit late, facilitated opportunities to discuss the future, end-of-life care and to make plans.

Families and carers matter even more

Data from family members came from two perspectives – both before and into bereavement. Many family members became informal caregivers. We found increased dependence on family members whether emotionally, physically, financially or for support with housing. The level of independence varied between the two age groups with those aged 16-24 more likely to be living in the parental home, still in education or receiving training and moving towards becoming independent from their family. Those aged 25-40 were more likely to have been independent adults for longer. In common with Knox et al, we also found that when thrust back into dependent relationships with parents, left behind by peers, whom they perceived to be moving forward with their own life goals, young adults could feel isolated.(26) The financial burden of cancer is widespread, but for those at the younger end of the age group who were still in education or living at home, the burden fell more heavily on their family. For those with greater independence and who relied on their income from employment, a cancer diagnosis compromised their ability to work and maintain their lifestyle. It is likely that older people with cancer, particularly those who have retired with an income to cover their regular expenses may not face such financial extremes. Mohammed et al. refer to caregivers ‘taking charge’, thrust into a role for which they often felt ill-prepared.(27) In our study, lack of understanding of the clinical situation due to confidentiality, a lack of practical or technical knowledge or skills and poor information from HCPs, themselves often reluctant to undertake end-of-life discussions, were some of the contextual influences contributing to feeling ill-prepared, abandoned or distressed.

The burdens for healthcare professionals

Professionals reported difficulty addressing the needs of both the person with cancer and their family as often they had different expectations. Professionals were aware that providing bereavement support to a family was difficult if they had not built a relationship with them in the limited time available. This is mirrored by our finding that continuity mattered much more to young adults. Managing complex family dynamics was challenging for HCPs and strategies to do this were often not addressed. Sometimes HCPs did not want to 'open a can of worms' by involving the family as they were aware that they would have to consider extra care needs, not viewed as part of their role. This was a strategy used by HCPs to manage their workload and families were not told that it was acceptable to ask for help and support. Professionals preferred to maintain and share optimism with the family, maintaining hope, all of which helped to reduce the emotional discomfort they would otherwise feel. So, talking openly about the death of the person with cancer was rarely pursued. Beerbower et al. refer to 'a broken system of communication' that can lead to conflict, where there has been no disclosure of prognosis, or where disclosure has for some family members only been partial, or come much too late.(20, 28) Educating, enabling and supporting caregivers can thus be complex and challenging, reinforcing the need for early and developmentally appropriate communication.

Professionals often have less exposure to and experience of providing end-of-life care for young adults. They are likely to be similar in age to the person with cancer, their family or friends, enhancing the emotional difficulties of working with this population. Whilst in palliative care, end-of-life might be 'normal', caring for those aged 16-40 who are dying will not be normal nor will facing the loss of lives partially lived. The avoidance by HCPs of engaging in the challenging discussions and activities we have already listed is understandable. But Wiener et al. point out that HCPs need to reflect and be aware of the emotional effect that younger patients have upon them and whether the support they offer is relevant and enabling of this population to continue to live normally for as long as possible.(29) Clark et al. have suggested that providing a developmentally-appropriate approach to care that includes advance decision making is thus essential.(30) To enable HCPs to meet the needs of the end-of-life care of young adults, formal support is needed. However, the formal support for HCPs in their professional roles varied in availability, access and was used differently. There was a distinction between doctors and nurses. Participants in our study suggested that the emotional burden received greater recognition in the nursing profession whereas for senior doctors there was little or no provision of support and an expectation that they would not show the emotional effect of their work. There were also issues about having the time to access support, associated costs and the lack of visibility and advocacy from senior HCPs for accessing support. In addition, support was not integrated into training or ongoing professional practice and for some senior HCPs it may have been regarded as compromising their role or authority.(31) Self-care in the palliative care workforce is known to be essential, yet rarely is education or training available.(32, 33) We would agree with Knox et al that palliative care services should consider prioritising resources to support self-care practice, to promote the health and well-being of HCPs.(18)

Strengths, limitations and future research directions

Although our study is unusual for the extensive data collected from young adults facing end-of-life and their triangulation with family and HCPs, recruitment of two groups of patients was unsatisfactory. Young adults with haematological malignancies were rarely invited to participate despite these being a commoner diagnosis in this population. This may be because those with haematological diagnoses continue to be offered and agree to receive ‘curative’ treatments.(34) When such curative options had been exhausted our participants were often ’actively dying’ and too ill to participate in this study. Another under-represented group were those aged 16-24. Professionals suggested that whilst clinical teams identified young adults meeting the study eligibility criteria, the challenges of communicating that ‘cure was not likely’ may have impacted on their reluctance to introduce the study. Our original plan to undertake two interviews with participants failed: often patients were just too unwell for a second interview. We cannot be certain that the data presented in this paper wholly reflects the experiences of these two populations, neither can we be certain of ‘completeness’ or ‘informational redundancy’, in these accounts; we are however more certain that ‘conceptual depth’ was reached. (35) Further research is needed to explore the needs of those often described as ‘hard to reach’, and those with haematological cancers and those aged 16-24 years. A further limitation arises from the recognised difficulties in estimating life expectancy so that study participants could not be accurately assessed as being within the last year of life and so some caution about their representativeness is necessary.

Conclusion

We identified challenges with the way current end-of-life care is delivered to young adults with cancer. Using this evidence, recommendations to improve care can now be developed.

References

1. Bleyer A, Ferrari A, Whelan J, Barr RD. Global assessment of cancer incidence and survival in adolescents and young adults. Pediatr Blood Cancer. 2017;64(9).
2. What are the top causes of death by age and gender? [Internet]. 2012 [cited 02 August 2016]. Available from: http://webarchive.nationalarchives.gov.uk/20160105221750/http://www.ons.gov.uk/ons/rel/vsob1/mortality-statistics--deaths-registered-in-england-and-wales--series-dr-/2012/sty-causes-of-death.html.
3. Globocan 2012: Estimated Cancer Incidence, Mortality and Prevalence Worldwide in 2012 [Internet]. 2012 [cited 02 August 2017]. Available from: http://globocan.iarc.fr/Pages/fact_sheets_population.aspx.
4. Rosenberg AR, Wolfe J. Palliative care for adolescents and young adults with cancer. Clin Oncol Adolesc Young Adults. 2013;2013(3):41-8.
5. Ngwenya N, Kenten C, Jones L, Gibson F, Pearce S, Flatley M, et al. Experiences and Preferences for End-of-Life Care for Young Adults with Cancer and Their Informal Carers: A Narrative Synthesis. J Adolesc Young Adult Oncol. 2017;6(2):200-12.
6. Bleyer A, Barr R, Ries L, Whelan J, Ferrari A, editors. Cancer in Adolescents and Young Adults. 2nd ed. Switzerland: Springer; 2017.
7. Grinyer A. Palliative and End of Life Care for Children and Young People: Home, Hospice, Hospital. Oxford: Wiley; 2012.
8. Grinyer A, Thomas C. The importance of place of death in young adults with cancer. . Mortality. 2004;9(9):114-31.
9. Kars MC, Grypdonck MH, de Bock LC, van Delden JJ. The parents' ability to attend to the "voice of their child" with incurable cancer during the palliative phase. Health Psychol. 2015;34(4):446-52.
10. Kars MC, Grypdonck MH, de Korte-Verhoef MC, Kamps WA, Meijer-van den Bergh EM, Verkerk MA, et al. Parental experience at the end-of-life in children with cancer: 'preservation' and 'letting go' in relation to loss. Support Care Cancer. 2011;19(1):27-35.
11. Royal College of Physicians Our Future health. Talking about dying: How to begin honest conversations about what lies ahead. London: Royal College of Physicians; 2018 October 2018.
12. Baker JN, Hinds PS, Spunt SL, Barfield RC, Allen C, Powell BC, et al. Integration of palliative care practices into the ongoing care of children with cancer: individualized care planning and coordination. Pediatr Clin North Am. 2008;55(1):223-50, xii.
13. Grinyer A, Barbarachild Z. Teenage and young adult palliative and end of life care service evaluation. London; 2011.
14. Pawson R, Tilley N. Realistic evaluation. London: Sage Publications Ltd; 1997. 235 ; 23 cm. p.
15. The Rameses II Project. “Theory” in realist evaluation 2017 [Available from: http://ramesesproject.org/pgm-download_media.php?name=RAMESES_II_Theory_in_realist_evaluation.pdf.
16. The Rameses II Project. Developing Realist Programme Theories 2017 [Available from: http://ramesesproject.org/pgm-download_media.php?name=RAMESES_II_Developing_realist_programme_theories.pdf.
17. Wong G, Westhorp G, Manzano A, Greenhalgh J, Jagosh J, Greenhalgh T. RAMESES II reporting standards for realist evaluations. BMC Med. 2016;14(1):96.
18. Richards L, Qualitative S, Research. Using NVivo in qualitative research. London: Sage; 1999. ix, 218 pages : illustrations ; 25 cm. + 1 CD-ROM (4 3/4 in.) p.
19. Charmaz K. Grounded Theory: objectivist and constructivist methods In: Denzin NK, Lincoln YS, editors. Handbook of qualitative research. California: Sage Publications Ltd; 2000. p. 1065 ; 26 cm.

20. Beerbower E, Winters D, Kondrat D. Bio-psycho-social-spiritual needs of adolescents and young adults with life-threatening illnesses: Implications for social work practice. Soc Work Health Care. 2018;57(4):250-66.
21. Soanes L, Gibson F. Protecting an adult identity: A grounded theory of supportive care for young adults recently diagnosed with cancer. Int J Nurs Stud. 2018;81:40-8.
22. Livneh H. Psychosocial Adaptation to Cancer: The Role of Coping Strategies. Journal of Rehabilitation. 2000;66(6):40-50.
23. Nissim R, Rennie D, Fleming S, Hales S, Gagliese L, Rodin G. Goals set in the land of the living/dying: a longitudinal study of patients living with advanced cancer. Death Stud. 2012;36(4):360-90.
24. Bell CJ, Zimet GD, Hinds PS, Broome ME, McDaniel AM, Mays RM, et al. Refinement of a Conceptual Model for Adolescent Readiness to Engage in End-of-Life Discussions. Cancer Nurs. 2018;41(2):E21-E39.
25. Llewellyn H, Neerkin J, Thorne L, Wilson E, Jones L, Sampson EL, et al. Social and structural conditions for the avoidance of advance care planning in neuro-oncology: a qualitative study. BMJ Open. 2018;8(1):e019057.
26. Knox MK, Hales S, Nissim R, Jung J, Lo C, Zimmermann C, et al. Lost and stranded: the experience of younger adults with advanced cancer. Support Care Cancer. 2017;25(2):399-407.
27. Mohammed S, Swami N, Pope A, Rodin G, Hannon B, Nissim R, et al. "I didn't want to be in charge and yet I was": Bereaved caregivers' accounts of providing home care for family members with advanced cancer. Psychooncology. 2018;27(4):1229-36.
28. Bell CJ, Skiles J, Pradhan K, Champion VL. End-of-life experiences in adolescents dying with cancer. Support Care Cancer. 2010;18(7):827-35.
29. Wiener L, Weaver MS, Bell CJ, Sansom-Daly UM. Threading the cloak: palliative care education for care providers of adolescents and young adults with cancer. Clin Oncol Adolesc Young Adults. 2015;5:1-18.
30. Clark JK, Fasciano K. Young adult palliative care: challenges and opportunities. Am J Hosp Palliat Care. 2015;32(1):101-11.
31. Bradford NK, Greenslade R, Edwards RM, Orford R, Roach J, Henney R. Educational Needs of Health Professionals Caring for Adolescents and Young Adults with Cancer. J Adolesc Young Adult Oncol. 2018.
32. Mills J, Wand T, Fraser JA. Self-Care in Palliative Care Nursing and Medical Professionals: A Cross-Sectional Survey. J Palliat Med. 2017;20(6):625-30.
33. Mills J, Wand T, Fraser JA. Examining self-care, self-compassion and compassion for others: a cross-sectional survey of palliative care nurses and doctors. Int J Palliat Nurs. 2018;24(1):4-11.
34. Odejide OO, Cronin AM, Earle CC, Tulsky JA, Abel GA. Why are patients with blood cancers more likely to die without hospice? Cancer. 2017;123(17):3377-84.
35. Saunders B, Sim J, Kingstone T, Baker S, Waterfield J, Bartlam B, et al. Saturation in qualitative research: exploring its conceptualization and operationalization. Qual Quant. 2018;52(4):1893-907.

Legends for figures

Figure 1. Phases of research process. Phases ii-iv are reported here.

Figure 2. Analysis process

Figure 1. Phases of research process. Phases ii-iv are reported here

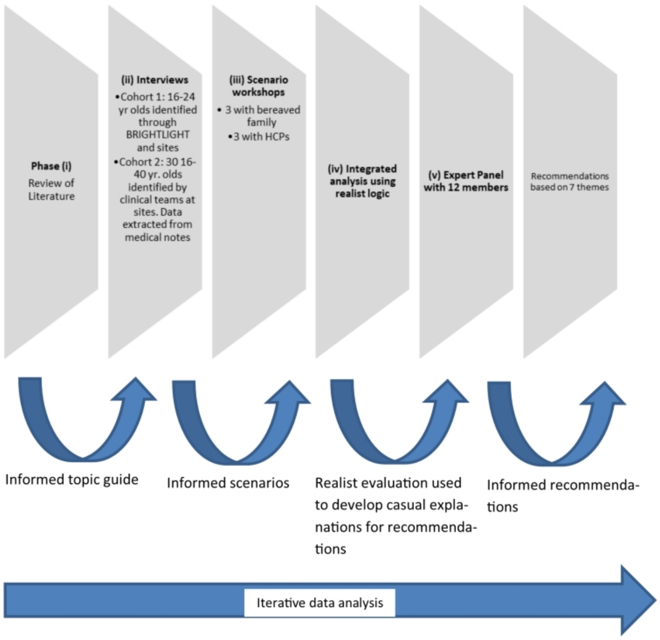
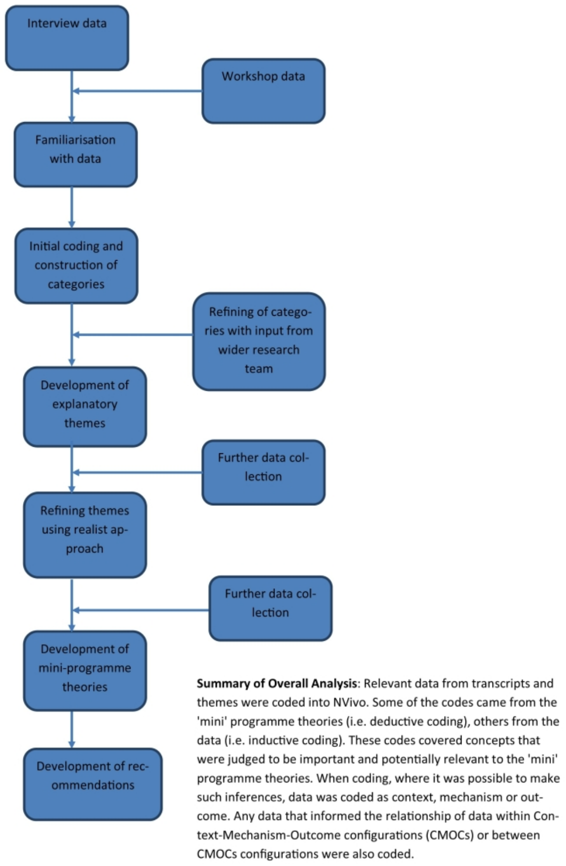


Figure 1. Phases of research process. Phases ii-iv are reported here.

209x297mm (300 x 300 DPI)

Figure 2. Analysis process



Summary of Overall Analysis: Relevant data from transcripts and themes were coded into NVivo. Some of the codes came from the 'mini' programme theories (i.e. deductive coding), others from the data (i.e. inductive coding). These codes covered concepts that were judged to be important and potentially relevant to the 'mini' programme theories. When coding, where it was possible to make such inferences, data was coded as context, mechanism or outcome. Any data that informed the relationship of data within Context-Mechanism-Outcome configurations (CMOCs) or between CMOCs configurations were also coded.

Figure 2 Analysis Process

288x297mm (300 x 300 DPI)

University College London Hospitals NHS
NHS Foundation Trust

Title of project: When cure is not likely: What do young adults with cancer and their families need and how can it best be delivered? A BRIGHTLIGHT companion study

PROTOCOL
Version 2.0 19th December 2013

UCL

Protocol Authorisation

PRINCIPAL INVESTIGATOR

Print name …………………………………………………………..

Signature …………………………………………………………..

Date …………………………………………………………..

When cure is not likely

Amendment History

Version	Date	Author	Application reference	Notes

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Lay Abstract

Background

This study will help increase our knowledge to understand better the needs of 16-40 year olds (“young adults”) when cure from cancer becomes unlikely. The younger members of this group are on the borders of paediatric care and as cancer is relatively uncommon in this age group, patients are less often encountered in adult cancer and palliative care services. Overall cancer is relatively uncommon in people in early to mid-adulthood. Nevertheless, one quarter of deaths in 16-40 year olds are due to cancer. The impact of incurable cancer on patients and families is strongly influenced by age but for adults in early to mid-life, very little is known about their experiences as death approaches or how care is best delivered. Professionals consistently acknowledge many specific challenges of managing end of life care in this age group.

From this study we aim to understand;

a) The most important parts of care in the last year of life for people with cancer aged 16-40 years.
b) Whether differences exist between the experiences of people with cancer who are aged 16-24 and those aged 25-40 years.
c) How young adults and their families can be supported in the last year of life to achieve their preferences for care.
d) The challenges that exist for health and social care professionals providing care.

Methods

There are five distinct parts to our research.

I. **Analysing available information:** we will look at all available literature and information about end of life care in young adults including the information that is now collected routinely in the NHS about preferences and place of death.
II. **Interviews with patients, families and professionals**. We will work closely with health care professionals from four study sites (University College London Hospitals, Southampton University Hospitals, Leeds Teaching Hospitals, and St. Joseph’s Hospice) to identify patients for interview about their care when cure is no longer likely. Fifteen 16-24 year olds taking part in the BRIGHTLIGHT study (a study already underway in the UK) and thirty 16-40 year olds from the four study sites will be invited to participate. We will ask them to propose a family member or carer and their key worker to be interviewed as well.
III. **Workshops with patients and professionals.** We will conduct nine workshops for interactive discussion and to increase our understanding of the range of perspectives, opinions and experience. Three workshops will be with family members and carers, three with professionals and three bringing together family members and carers and professionals.
IV. **Hold professional panels.** We will present our findings and recommendations to a panel of professional experts for further refinement.
V. **Make recommendations for practice.** We will have a good understanding of the core components and pathways of end of life care for young adults and make recommendations for practice and further evaluation to assist policy makers, commissioners and other stakeholders.

How the results of this study will be used

The findings will add evidence to inform national standards, pathways of care and core competencies for training staff. We will identify areas for change or further exploration. We will make public our findings widely to both lay and professional audiences.

1. Purpose of the study

This research will illuminate the core issues affecting end of life care[1] in young adults with cancer (aged 16-40 years), gathering evidence from the perspectives of the young people themselves, their families and the multidisciplinary team.

Using a national, multiple method realistic evaluation, we shall use an iterative approach guided by the MRC framework for evaluating complex interventions. [1,2] The aim is to define, describe and understand the core components for excellent practice in the delivery of end of life care for young people with cancer, to inform policy and practice and to set priorities for further evaluation studies.

In the United Kingdom (UK), health policy on specialist cancer services has bracketed young adults up to 24 years with teenagers (teenagers and young adults, TYA). [3] In the United States, Canada and Australia, strategies for improving cancer outcomes have most often focussed on 15-40 year olds ('adolescents and young adults', 'AYA'). [4] Notably, Douglas House, a unique hospice for young adults in Oxford serves an age range of 16-35 years. [5] This age group has attracted little attention in other European countries where there has been less consistency in the age range studied. We aim to explore comparisons between those aged 16-24 years and those aged 25-40 years to highlight key issues and differences that may be influenced by age.

In devising this research study, our underlying theories are:

1. That end of life care for young adults with cancer aged 16-40 years could be improved by increased understanding of (a) current care pathways for people with different types of cancer (b) the effects of age (above and below 25 years) (c) the need for accurate information (d) how active participation by young adults in decision making can impact on current and future care (e) the importance of respect for individual autonomy and family interactions.

2. That outcomes would be improved by an approach to end of life care that is (a) more aware of individual patient need and autonomy (b) supports professionals to recognise and respond to patient need (c) takes account of family interactions and relationships with patient and staff (d) enables patients to receive active and palliative treatments in a place of their choice to achieve their preferences for end of life care.

Thus, our objectives are to understand:

a) The core components in the pathways of care in the last year of life for people with cancer aged 16-40 years.
b) Any differences between the experiences of people with cancer from the age ranges 16-24 and 25-40 years.
c) How young adults and their families can be supported in the last year of life to achieve their preferences for care.
d) The challenges that exist for health and social care professionals providing care.

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[1] End of life care is defined as 'care that helps all those with advanced, progressive, incurable illness to live as well as possible until they die'. [6]

2. Background

One quarter of deaths in 16-40 year olds are related to cancer.[7] Overall survival rates have improved less than those of younger children and older adults.[8] Despite descriptions of the specific needs of teenagers and young adults for specialist cancer care, there is a dearth of empirical research, policy and expert practice related to their End of Life care. This is also true for young adults up to 40 years.

There are gaps in policy for this age group. Better care: Better Lives [9] makes no distinction between the needs of children and teenagers and young adults and deals exclusively with children's palliative services. Similarly the End of Life Care Strategy, Promoting High Quality Care For All Adults At The End Of Life makes no specific reference to young adults.[10]

Published literature about End of Life for teenagers and young adults has been confined to summaries of good practice or, where studies have been undertaken, data has been sourced from parents rather than young people themselves.[11-13] Others have undertaken retrospective analysis of medical notes [14] or produced comment and review papers.[15-18] Notably, just one study interviewed young people and explored their views on decision making as End of Life approached.[19]

Equally, work on the older young adult cancer population is limited. The few studies that have collected data from adults of all ages, either through interviews or questionnaires (EORTC QLQ-C30) have identified differences between ages at end of life including varying preferences for active treatment and greater symptom burden or lower quality of life.[20-22]

For those aged below 40 years, boundaries between curative and palliative treatment are often blurred; decisions may be influenced by cancer type, age and family circumstances. There is a lack of standardized models, approaches and communication aids for this age group; the management of symptoms and psychosocial concerns may be neglected if the challenges to communication are not overcome. Avoidance and delaying of discussions about End of Life decisions are common resulting in consequences for the quality of care.[15]

The challenges in delivering care to young people are as relevant to the delivery of end of life care as to intensive curative chemotherapy.[23-24] National policy in England and Wales directs that TYA have access to specialist services to meet specific challenges faced by young people in accessing services responsive to needs associated with the physical, educational, psychological and social developmental stages that are disrupted by the diagnosis and treatment of cancer.[4] This policy is being evaluated in the BRIGHTLIGHT study, (NIHR RP-PG-1209-10013; Appendix 3, study synopsis).

BRIGHTLIGHT will provide: a cohort of participants of all those aged 13-24 diagnosed with cancer in England in one year; the contexts and pathways of cancer care for young adults in England; sources of contributors to workshops and expert panels; prospectively collected patient outcome data and extensive analysis of the national cancer and end of life data sets. This takes into account information already available in the national cancer data repository (NCDR) and from the National End of Life Care intelligence network (Nend of lifeCIN) for those aged 16-40 years. BRIGHTLIGHT and this proposal are closely linked to the remit of the National Cancer Research Institute Teenage and Young Adult Clinical Studies Group.[25]

This study will provide the much-needed evidence to underpin quality of care and the enablers that will assist in addressing current barriers:

	Barriers to excellent end of life care	**Enablers of excellent end of life care**

We can influence	Evidence base: Poor understanding of (a) disease trajectory; (b) unmet health and social care needs; (c) variations between cancers; (d) variations between age, groups, gender, ethnicity Service factors: Support needs of staff , patients and families in decision making Attitudes and barriers to care of young people: structural; cultural; financial. Individual factors: personal and disease specific, education Costs of care	Data from BRIGHTLIGHT, NCDR and Nend of lifeIN Data from workshops and interviews Training and support health and social care professionals Understanding of the costs of components of end of life care components
What will influence our project	Recognition of last year of life and end stage disease Interaction between active and palliative treatments Family dynamics Attitudes of health care professionals National factors in healthcare environment; costs of care (perceived and actual), commissioning Regional variation in policy, service configuration and provision; cultural and economic factors Training and support needs of health and social care professionals	Integrated care pathways Strategic documents; DH and voluntary sector pressures; new service providers; public opinion; epidemiological trends Assessing transferability of intervention in regional sites Engagement of clinical champions; multi-disciplinary team approach to care Training and support for health and social care professionals

3. Study Design and Methods

Research will be undertaken in 5 discrete but inter-related phases.

(i) Preparatory phase;
(ii) Interviews with young adults, families and professionals;
(iii) Scenario workshops with families and professionals;
(iv) Analysis and interpretation of these data including synthesis with a literature review and quantitative data available through NCDR, Nend of lifeCIN and BRIGHTLIGHT then refinement through discussion with a panel of experts;
(v) Development of recommendations for practice and further evaluation.

See Appendix 1 for diagram of these phases.

(i) Preparatory phase

This will be undertaken at UCLH and includes a detailed synthesis of the literature relevant to end of life for young adults with particular emphasis on identification and understanding of the mechanisms potentially causing the desired outcomes. Additionally, patterns of care described by National Cancer Data Repository and National End of Life Care Intelligence Network will be used to give further characterisation by definition of tumour types responsible for deaths, place of death and variations within our age range. This phase will inform the semi-structured realist interviews with young adults and scenario development.

(ii) Interviews

Several groups including patients, families and professionals will be interviewed as described below. Four sites will be involved in this part of the study.

We will conduct interviews with 45 young adults aged 16-40 years of age, purposively sampled to reflect a range of diagnoses (relevant groupings include leukaemia, lymphoma and solid tumours including brain, testis, sarcoma, carcinomas). Recruitment will be from the two sources outlined below (Appendix 2 illustrates recruitment, consent and data collection processes).

Sample 1 will be 15 teenagers and young adults aged 16-24 years participating in the BRIGHTLIGHT[2] cohort who will ‘self-identify’ through responses to trigger questions in the BRIGHTLIGHT survey indicating that a) no more treatment is possible, b) they have been offered/received care from the symptom control team or palliative care team or c) giving a response to the question asking what they had been told about their cancer suggesting that they are aware that cure is no longer likely. The BRIGHTLIGHT Senior Research Manager (SRM) or Cohort Manager (CM) will confirm with the young person’s healthcare team that they are receiving end-of-life care (if this was not confirmed on the pre-survey check) and make sure that it is suitable to approach them to take part in the study. When this is assured they will call the young person, briefly describe the study and gain verbal consent to forward the information sheet. After approximately a week, the young person will be contacted via an appropriate means (e.g. ‘phone, text) by the same person (SRM or CM) to see if they have received the information sheet and asked if they would like to take part in the study. For those who agree they will be asked for verbal consent to pass their contact details onto the RA who will then contact the young person.

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[2] See www.brightlightstudy.com for details of BRIGHTLIGHT and appendix 3 for a synopsis.

If a young person verbally consents to participate the RA will arrange a time and place to meet them for the interview. On the day before the interview the RA will call to confirm the young person would still like to participate; if they do then the RA will visit the young person as planned but before commencing the interview, s/he will confirm the young person understands what they are agreeing to and get written consent. For a detailed description of this recruitment process, refer to Appendix 4.

After a first interview, this group will be invited to participate in a second interview between 2 and 4 months later. At the time when the young person is recruited, they will be asked to nominate a key worker who will be in charge of providing support during the study. This key worker will be in contact with the researchers and will follow up with the young person after the interview to see if they need additional support.

Sample 2 will be interviews to include thirty patients aged 16-40 years. Recruitment will be facilitated by clinical staff at six study sites: University College London Hospitals, Southampton University Hospitals, Leeds Teaching Hospitals, St Gemma's Hospice, Wheatfields Hospice and St. Joseph's Hospice. These research sites were selected because they all have established palliative care teams and services, they actively participate in research, and the staff are skilled at recruiting patients in End of Life for research.

A poster will be displayed in appropriate spaces with contact details of the site PI and potential participants asked to talk to their clinician if they are interested. Once members of the clinical team have identified potential participants, they will make sure their details can be shared with the researchers. If the participant agrees, the researchers will contact them with more information about the study. Whenever possible, this will be done through a face to face conversation about the study, but if this is not possible, the information will be sent via post and questions will be answered over the telephone. The participants will then be given time to look at the information sheets and ask questions about the study. If the participant agrees to take part in the study, they will be asked to sign a consent form.

The young people in sample 2 will be asked to identify a family member, and a professional directly involved in their care such as the key worker who can be approached to be interviewed in addition (total 90 interviews). The young people will be asked to check with the family member to see if they agree that their details be passed to the researchers so that they can contact them with more information about the study. If the family member agrees, the researchers will contact them to talk about the study and provide them with the information sheet. The researchers will also approach the nominated healthcare professionals to provide them with more information about the study and to see if they are interested in taking part.

This second sample will allow targeting of patients identified by their key workers as being directly aware that cure is no longer likely and currently in the end of life phase. Ten will be aged 16-24, and 20 between 25-40 years. The sample size and distribution has been selected to add additional interviews to complement sample 1, to reflect the clinical heterogeneity present across this age range and in anticipation of saturation of themes during analysis (refer to Appendix 5 for the sampling matrix).

Using an investigator designed template (see Appendix 6), medical notes of interviewees will be reviewed to analyse written communication and documentation about discussions related to end of life in order to seek illustrations of best practice. Records of key discussions will be sought including communication between health professionals; record of advance care planning, do not resuscitate orders; communication about preferred place of death; insight of patient and family; information given and received; and evidence to show if limits of desired levels of information had been elicited. We will pilot the review of the notes at the beginning of the period of data collection and adjust the template to suit the content found in the medical notes.

All semi-structured realist interviews will be conducted by a researcher experienced in working with young adults and discussing sensitive issues. It is anticipated they will last for approximately one hour, will be digitally recorded, and transcribed verbatim prior to qualitative analysis of transcripts. Interviews will occur in the place of choice of participants, usually during clinic visits or in their own homes.

(iii) Workshops

Scenario development: end of life trajectories for young adults with cancer will be derived from an initial scenario development workshop attended by palliative care consultants, palliative care and cancer clinical nurse specialists, allied health professionals, oncologists and haematologists. Findings from the literature review, analysis of National Cancer Data Repository and National End of Life Care Intelligence Network data and emerging findings from the interview data will be presented. Scenarios will be co-constructed to precipitate discussion around key influencing variants such as diagnosis, different ages within the range being studied, symptom constellations and patterns of information disclosure and communication (for examples refer to Appendix 7).

Nine scenario workshops: A series of three workshops will be held in the study sites used in (ii) above using the scenarios to encourage interactive discussions and generate new thinking and potential solutions to problems raised. The use of different sites will support comparisons across contexts and address issues of generalisability and transferability of findings across different UK settings. Workshops at each site will be held with:

1. Families and carers of young adults with cancer; patient representatives and regional stakeholder organisation representatives (workshop maximum size 15)
2. All members of the multiprofessional team involved in delivery of treatment for cancer and end of life care (workshop maximum size 15)
3. Mixed participants representing groups (1) and (2) (workshop maximum size 20)

The family members and healthcare professionals will be recruited using two routes. First, from the participants who were interviewed as part of sample 2. The consent forms for the interviews explained in (ii) will include a section where the family members and healthcare professionals can indicate if they would like to be contacted to participate in the workshops. Second, additional family members and healthcare professionals will be recruited in the study sites by the researchers and members of the clinical team. Close contact with the clinical team will facilitate recruitment.

Consensus will not be sought, rather perspectives, opinions and experiences elicited allowing scenario ‘mapping’. This will be done in small groups using mapping aids to identify key ideas and hypotheses arising on how end of life care could be improved.

All workshops will be moderated by members of the research team who have experience of these methods as well as end of life/cancer care. Workshop teams will be developed to ensure consistency across this aspect of data collection.

(iv) Expert panel review

A panel of experts will be convened to include palliative care professionals, general practitioners, oncologists, haematologists, allied health professionals, policy makers, ethicists, charities and commissioners. Participants will not have been involved in previous workshops or interviews. Professionals will be identified by previous participants and members of the

research team. Professional organisations such as the NCRI Palliative and Supportive Care Clinical Studies Group will be consulted. These will be purposively selected based on their experience working with young people who are receiving end of life care, knowledge of policy issues and agree to participate.

A distillation of knowledge and understanding gained from realist interviews and workshops combined with quantitative data, will be presented to the expert panel describing ideal pathways with an explanatory account of key components of care. The panel will be asked to comment on areas of agreement and disagreement, provide alternative or additive explanations and to test and retest experientially the fit of mechanisms to potentially achievable outcomes as described in the pathways.

(v) Development of recommendations

The analysis and interpretation by an expert panel will lead to the writing and refining of an overarching explanatory account for end of life care for young adults with cancer. We shall present data to inform our objectives stated in section 1.

Emerging national standards, pathways of care and core competencies are anticipated and will be considered within the document. We shall circulate our draft to participants in the realist interviews and workshops, policy makers, commissioners and other stakeholders and take account of their comments in producing a final version. Identification of areas for potential intervention will be included and recommendations for future empirical evaluation and testing of the effect on outcomes will be suggested. Such empirical work would be the subject of the next phase in testing the feasibility and acceptability of the core components of delivering improved end of life care in a pilot observational study or exploratory randomised trial in the future. This would include consideration of the economic aspects of these approaches to care.

4. Data analysis

Methodological approach

We will use realistic evaluation, derived from critical realism. Its strengths are an emphasis on understanding the causal mechanisms which generate outcomes, consideration of context and a desire to improve practice and service delivery.[26] It supports a mixed method, iterative approach to capture multidimensional aspects of the evaluation of end of life care, which can be viewed as a complex, multi-component intervention. Realist research explores the links between context, mechanism and potential outcome. It increases our understanding of ‘what works, for whom, in what circumstances, in what respects and why?’ It seeks to penetrate beneath the observable inputs and outputs of an intervention. We shall initially untangle the influence of context in the care of young adults in age groups 16-24 and 25-40 years, with a range of cancer diagnoses, experiences and expectations of care. We shall use workshops to generate hypotheses on mechanisms by which care in the last year of life for people aged 16-40 years might be improved. Using qualitative data and quantitative data we shall assess which components of an intervention or approach to end of life care might lead to improvements in patient and family centred outcomes.

Analysis of interviews

Sample 1: Transcripts of the interviews will be analysed using a grounded theory approach. This provides a systematic and inductive approach for the collection of data, sampling and the building of theoretical frameworks.[27] Analysis will occur simultaneously with data collection. After reading and re-reading the transcripts, memoing and selective focused coding, constant comparison between codes will take place leading to development of categories. Software such as Atlas.Ti will support this. The second interviews facilitated by theoretical sampling will ensure completion of any conceptual gaps in the emerging theoretical framework with an understanding of individual experiences over time.

Sample 2: Grounded theory methods of analysis will also be used. The development of codes and constant comparison of codes in the development of categories will occur across the sources of data for each young adult. For example data will be triangulated using the interview data from the young person, their family member, nominated health professional and analysis of the medical notes. When a conceptual understanding has been developed for each case, analysis will occur across the sources of data as a whole.

Analysis of workshops

Detailed field notes will be taken, memos and post-it notes collected and workshop discussions will be audio-taped and transcribed verbatim as discussion around scenarios is as crucial as the 'mapping'.[28] Data will be entered into Atlas. ti. The research team will create a preliminary analysis from initial observations of the scenario maps developed and the transcripts of discussions from workshops (1) and (2) to generate a series of hypotheses of how end of life care can be improved. This initial analysis will be used to inform discussions in workshop (3).

After the data from the workshops is analysed, a short report will be distributed among the participants in order to share the findings as near to the time as data collection as possible.

Analysis for expert panel review

Analysis of data collected in an iterative process, a key aspect of realist methods, will be the focus. Preliminary thematic summaries of findings from the interviews and workshops will be combined with emerging quantitative data. Hypotheses on mechanisms of how end of life care could be improved will be carefully defined and prioritised. These will be refined further through discussion within the research team and with a panel of experts. This phase will allow a reconsideration of understanding of the interrelationships between the context and mechanisms generated by the experience of end of life care derived from all aspects of data collection, testing assumptions and exploring further remaining uncertainties.

5. Ethical considerations

We recognise that this is a sensitive area of research and we will be working with vulnerable participants. We anticipate the fact that the questions included in our interview could pose potential emotional and psychological burden for those involved. We are sensitive to this fact. The clinical members of our research team have significant experience in this area that we can draw upon. In addition we will make use of the now growing body of evidence of published studies on this topic.[29,30]

Therefore, we have taken the following measures in order to minimise the risks and burden for the research participants. The researchers to be appointed will have extensive experience of gaining consent and interviewing vulnerable participants. The researchers will ensure the protection and well-being of the participants throughout the entire duration of the study. The participants will be informed that they can ask questions or express their concerns about the study throughout its entire duration and can withdraw at any point. The researchers will also search for signs of discomfort or distress among the participants and will address them individually by talking to the participants and letting them know their options for withdrawal. During the discussion, the participants will be informed that they can refuse to answer questions. The information sheets contain the contact information of all of the members of the research team. The participants will be informed that they are free to contact the researchers with questions and concerns even after the study has ended.

Inbuilt support will be embedded in liaison with key members of the clinical team for recruitment, consent and support mechanisms for the participants if the interviews give rise to issues that need to be followed up (see Appendix 2). The researchers and the members of the clinical team will have ongoing communication about recruitment and data collection. The

members of the clinical team will play an instrumental role during the screening and selection process with the purpose of identifying potential participants and guaranteeing their protection. The key worker will be in charge of following up with the participant after the interview to see if any issues need to be discussed and provide appropriate guidance and assistance. This is important as the research may lead specific patients to face and discuss issues that had previously not been raised.

Considerable researcher burden is also possible. Having two researchers will ensure mutual support. There will also be extensive support from the research team, where expertise in methods and cancer care will be closely matched with researcher need and support. Monthly clinical supervision has been costed into our study.

6. User involvement

This aspect presents specific challenges and has been approached as follows: young adults and their families will be involved in data collection, feedback and analysis as an intrinsic aspect of our study; the NCRN Consumer Liaison Group has been approached to determine specific interest amongst members; the proposal will be read and commented on by family members of two young adults who died of cancer; input into patient information sheets and other patient information will be sought from the Young Peoples Reference Group associated with BRIGHTLIGHT (approximately 20 members - young people diagnosed with cancer between the ages of 14 and 25 (current age 18-29), some of whom have worked with us for over three years on various studies); additional input into patient and subject information sheets will be sought from an additional PPI group, the Cancer Partnership Research Group of the Surrey, Sussex and West Hampshire Clinical Research Network; presentations will be made from January 2013 to ‘Kayleigh’s workshop – terminally talkative’ at the annual young people’s conference, Find Your Sense of Tumour. This was first held in 2011 for those receiving end of life. Facilitated by two psychologists, it is now a regular event at the conference. The attendees from 2011 have also set up their own site on ‘Facebook’, moderated by psychologists, and which we can access if more immediate consultation on the study is needed. Through contact with the NCRN Consumer Liaison Group we have identified patients and patient representatives who will comment on the evolving study and contribute to workshops and panels.

We expect there will be very hard to reach participants consequent on limited awareness of End of Life, high levels of family and professional protection or exaggerated denial. Close relationships between researchers and key workers will be developed to improve chances of access. This will be enhanced by the clinical credibility and national profile of the research team.

7. Data sharing plan

BRIGHTLIGHT is approved by the National Information Governance Board reference ECC 8-05(d)/2011. Electronic data from the study will be stored on an NHS server supported by University College London Hospitals NHS Foundation Trust (UCLH). Paper documents will be stored in a locked filing cabinet in the cancer trials research facility at UCLH. Access will be limited to research associates and chief investigator. All digital recordings will be deleted once a written transcript has been produced. The interview and workshop transcripts will be anonymised and password protected. These will be archived, and available for further analysis after publication of the findings of BRIGHTLIGHT on End of Life.

8. Management of the study

The study is sponsored by UCL.
The core project team led by the CI work in close proximity and will meet regularly.
An advisory group will be established to regularly review and advise on study progress.

Timescales – total period 24 months

When cure is not likely

Month 1-4	Researchers start employment Detailed review of the literature and analysis of National Cancer Data Repository and National End of Life Care Intelligence Network Detailed protocol development
Month 5-17	Workshops and interviews
Month 18-21	Analysis
Month 21	Expert Panel
Month 22-24	Circulation of account to participants, policy makers, commissioners and other stakeholders. Completion of final account, dissemination through local meetings and writing of peer reviewed publications.

References

[1] Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M. Developing and evaluating complex interventions: the new Medical Research Council guidance. BMJ (Clinical research ed. 2008;337:a1655.
[2] MRC. Complex Interventions Guidance. 2008 [cited March 28th 2012]; Available from: (http://www.mrc.ac.uk/Utilities/Documentrecord/index.htm?d=MRC004871)
[3] National Institute of Health and Clinical Excellence. Improving Outcomes in Children and Young People with Cancer. 2005.
[4] Geiger AM, Castellino SM. Delineating the age ranges used to define adolescents and young adults. J Clin Oncol. 2011 Jun 1;29(16):e492-3.
[5] Helen and Douglas House. [cited 28th March 2012]; Available from: www.helenanddouglas.org.uk
[6] Department of Health. End of life care strategy. Promoting high quality care for all adults at the end of life. 2008.
[7] Office for National Statistics. Mortality Statisitics. 2010 [cited 28th March 2012]; Available from: http://www.ons.gov.uk/ons/publications/re-referencetables. html?edition=tcm%3A77-230730
[8] Barr RD. Adolescents, young adults, and cancer--the international challenge. Cancer. 2011 May 15;117(10 Suppl):2245-9.
[9] Department of Health. Better care: better lives. Improving outcomes and experiences for children, young people and their families living with life-limiting and life-threatening conditions 2008.
[10] Department of Health. End of life care strategy. Promoting high quality care for all adults at the end of life. 2008.
[11] Grinyer A, Thomas C. The importance of place of death in young adults with cancer. Mortality. 2004;9(2):114-31.
[12] Kars MC, Grypdonck MH, de Korte-Verhoef MC, Kamps WA, Meijer-van den Bergh EM, Verkerk MA, et al. Parental experience at the end-of-life in children with cancer: 'preservation' and 'letting go' in relation to loss. Support Care Cancer. 2009 Jan;19(1):27-35.
[13] Grinyer A. Palliative and End of Life Care for Children and Young People: Home, Hospice, Hospital. Oxford: John Wiley and Sons 2012.
[14] Cohen-Gogo S, Marioni G, Laurent S, Gaspar N, Semeraro M, Gabolde M, et al. End of life care in adolescents and young adults with cancer: experience of the adolescent unit of the Institut Gustave Roussy. Eur J Cancer. 2011 Dec;47(18):2735-41.
[15] Baker JN, Hinds PS, Spunt SL, Barfield RC, Allen C, Powell BC, et al. Integration of palliative care practices into the ongoing care of children with cancer: individualized care planning and coordination. Pediatric clinics of North America. 2008 Feb;55(1):223-50, xii.
[16] Christenson K, Lybrand SA, Hubbard CR, Hubble RA, Ahsens L, Black P. Including the perspective of the adolescent in palliative care preferences. J Pediatr Health Care. 2010 Sep-Oct;24(5):286-91.
[17] Pritchard S, Cuvelier G, Harlos M, Barr R. Palliative care in adolescents and young adults with cancer. Cancer. 2011 May 15;117(10 Suppl):2323-8.
[18] Wein S, Pery S, Zer A. Role of palliative care in adolescent and young adult oncology. J Clin Oncol. 2010 Nov 10;28(32):4819-24.
[19] Hinds PS, Oakes LL, Hicks J, Anghelescu DL. End-of-life care for children and adolescents. Seminars in oncology nursing. 2005 Feb;21(1):53-62.
[20] Lundh Hagelin C, Seiger A, Furst CJ. Quality of life in terminal care—with special reference to age, gender and marital status. Support Care Cancer. 2006 Apr;14(4):320-8.
[21] Parr JD, Zhang B, Nilsson ME, Wright A, Balboni T, Duthie E, et al. The influence of age on the likelihood of receiving end-of-life care consistent with patient treatment preferences. Journal of palliative medicine. 2010 Jun;13(6):719-26.
[22] Trevino KM, Maciejewski PK, Fasciano K, Greer JA, Partridge A, Kacel EL, et al. Coping and psychological distress in young adults with advanced cancer. Journal of Supportive

Oncology. 2011;http://www.supportiveoncology.net/single-view/aipcoping-and-psychological-distress-in-young-adults-with-advancedcancer/8ea0588008.html.
[23] Hollis R, Morgan S. The adolescent with cancer--at the edge of no-man's land. The lancet oncology. 2001 Jan;2(1):43-8.
[24] Whelan J. Where should teenagers with cancer be treated? Eur J Cancer. 2003 Dec;39(18):2573-8.
[25] National Cancer Research Network.
[26] Pawson R, Tilley N. Realistic Evaluation. London: Sage 1997.
[27] Charmaz K. Grounded Theory: objectivist and constructivist methods In: Denzin N, Lincoln Y, eds. The Handbook of Qualitative Research. 2nd ed. Thousand Oaks: Sage 2000:509-36.
[28] Oreszcyzn S, Carr S. Improving the link between policy research and practice: using a scenario workshop as a qualitative research toll in the case of genetically modified crops. Qualitative Research. 2008;8:473-97.
[29] Hinds PS, Burghen EA, Pritchard M. Conducting End-of-Life Studies in Pediatric Oncology. WJNR 29(4):448-465; 2007.
[30] Walshe CE, Caress AL, Chew-Graham C, Todd C. Case Studies: A Research Strategy Appropriate for Palliative Care? Palliative Medicine 18(8):677-684; 2004.

When cure is not likely

Appendices

Appendix 1: Phases of research
Appendix 2: Recruitment and data collection
Appendix 3: BRIGHTLIGHT study synopsis
Appendix 4: Sample 1 recruitment
Appendix 5: Sampling
Appendix 6: Review of medical notes
Appendix 7: Hypothetical scenarios

When Cure Is Not Likely - Methodology Protocol

Interviews and Workshops

[REC Ref: 13/LO/1098]

Rationale

This mini protocol outlines the approach to be implemented for the data collection phase (interviews, workshops and analysis). This will be informed by a realist approach which aims to understand the causal mechanisms which generate outcomes, consideration of context and a desire to improve practice and service delivery. Its iterative approach assists the capture of multidimensional aspects of the evaluation of End of Life Care and explores links between context, mechanism and outcome. It will increase our understanding of 'what works, for whom, in what circumstances, in what respects and why?'

WCINL: Aims and Objectives

Methods

Interviews

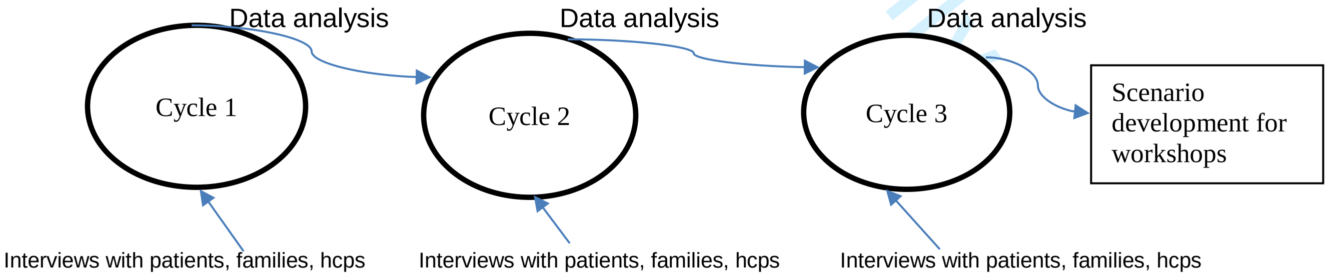
Realistic evaluation will be used as a guiding framework in exploring participants' experiences on the following outcomes;

Good patient centered care
Good family centered care
Responsive clinical care

Our understanding of what is implied by the term 'good care' will develop as our interviews take place and are analysed. For example, emotional spiritual physical; communication, information giving, management of symptoms; promotion of health family systems; social situation; quality of life; supporting finding meaning finding balance between acceptance and hope.

We plan a set of iterative cycles of interviews conducted with 3 groups in each cycle: patients, nominated family members and nominated health care professionals. We shall sample a maximum of 5 patients within each of the two age ranges (16-25 and 25-40) in each cycle, thus a maximum of 15 interviews per age group. Analyses of data will occur after each cycle and findings will be used to inform the conduct of the next cycle of interviews. Once data saturation is reached, the findings will be used to develop scenarios for use in the workshops in the next phase of our work.

Schema of iterative approach for interviews at Time 1:



The interviews will be explorative in their approach due to a significant lack of available evidence about the experiences of this population when cure is not likely and death approaches. This includes a lack of evidence on the availability and appropriateness of palliative approaches to care either alongside treatment or in the dying phase. Through the interviews we aim to explore and understand past and present experiences leading to how the future is viewed and conceptualized. We shall consider the internal, external and reflexive processes of the individual's experience. This will involve the different levels that impact on care: individual,

group (family and within healthcare professional teams), systemic and organizational (Ferlie and Shortell, 2001). We shall also explore the situational and contextual attributes for the individual.

The interviews will be iterative in their style using key questions and prompts to explore the perspectives and experiences of the participant. It is anticipated that each interview will last c. 1 hour. Interview schedules can be found at the end of this document, but in short;

> **Patients** will be asked to discuss their care, and perspectives of support for them, using the broad framework "What has happened, what is happening now, and what do you think will happen next?" Experiences of Internal processes; interpersonal and communication, relationship to others and the situation and context will be explored.
>
> **Family members** (nominated by patients) will be asked about the patient's experiences as well as their experiences and perspectives of support for themselves as a family and/those in a caregiving role. This will include considering the effects on family dynamics, communication between family/ young person and professionals; how families can be supported to enhance their capacity to manage patient care and their perspectives of the past, present and future needs of the patient. Family centered outcomes in loss include experiences in bereavement [could use bereavement risk assessment tools for guidance (Agnew et al., 2010)].
>
> **HCPs** will be asked about their experiences of providing care for the specific patient who nominated them, their more general experiences of caring for patients in this age group when cure is no longer likely leading to discussions to inform our understanding of their internal processes of dealing/coping with younger patients who face death. Here the realist approach will allow both interviewer and interviewee to contribute to discussions of working in an area where sensitive issues are commonplace, thus facilitating opportunities for greater insights.

Patient Age Range: 16-25 years
Cohort 1: BRIGHTLIGHT
Sample size: 15 patients over 3 iterative cycles (maximum 5 patients per cycle)
Sampled from 4 cancer groups – minimum of 3 patients from each group

- leukaemia, lymphoma
- bone and soft tissue sarcoma
- brain and CNS tumours
- carcinomas including germ cell tumours

Interviews in this cohort will take place at time 1, and again after 2-4 months (time 2). The second interviews will explore the current situation and what has changed for the patient since the first interview using the same interview schedule but with a greater focus on what has changed. We anticipate that patients will have experienced both external and internal changes. We shall explore how they have experienced changes in their care, their relationships, and how they now conceptualise the future.

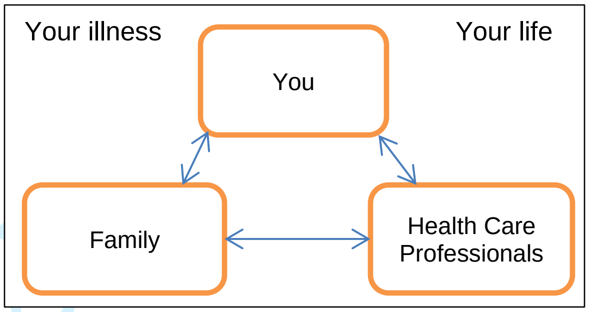
Patient age ranges: 10 16-24 yr olds; 20 25-40 yr olds
Cohort 2: Sites
Sample size: 30: interviews will occur at time 1 only.
Four cancer groups:

- Any carcinomas including breast, colorectal, melanoma, gynaecological and rare tumours – the most frequent diagnoses, so sample to recruit minimum of 21 patients
- Bone and soft tissue sarcoma – minimum 3
- Brain and CNS tumours – minimum 3
- Leukaemia, lymphoma – minimum 3

Conduct of semi-structured realist interviews:
A realist interview allows both interviewer and interviewee to contribute to discussions to facilitate deeper exploration of pertinent issues and allow the possibility of considering innovative issues and solutions.

Interviewers will work to a simple topic guide, allowing free discussion of issues in the past and present and what may occur in the future.

If a patient has difficultly answering questions or conveying their experiences a simple visual prompt may be used (example below) to provide a focus



Simple visual interview prompt

Analysis
All interviews will be audio-taped and transcribed verbatim. Two members of the research team will read in depth each transcript.

A modified grounded theory approach to analysis will be used (Charmaz, 2006). This will take an interpretative approach to identify themes and look for meanings and relationships within the interview data.

Each patient (cohort 2) will generate a set of 3 interviews which will be considered together as case studies which will then be compared and contrasted. In addition, the full set of each category of interviews (patient, family and HCP) will be analysed separately to extract common themes. We shall compare the datasets for the two age groups and look for commonalities and differences. Our findings will be used to inform the development of scenarios to be used in the workshops in the next phase of our research.

We shall also use our findings to develop further our programme theories. These theories have begun with the underlying hypotheses and enablers and barriers that we have identified at the start of our work from the literature and expert opinion of the research team. By defining our proposed outcomes as the provision of ‘good’ experiences of care for patients and families, and responsive care from HCPs, we shall use our data to develop a fuller understanding of what constitutes good and responsive care, and good experiences for patients and families. We shall use diagrams, flow charts, maps and memos to develop a full picture.

Outline of next stage of research:
Scenario workshops
Workshops will be held across our three recruitment sites across UK, Leeds, London and Southampton. Three workshops will be held at each site (total of 9) comprising of 1 with family members; 1 with healthcare professionals and 1 mixed family and HCP. The scenarios will be used to facilitate interactive discussion of issues arising for patients, families and HCPs in the experiences of care and illness for people in the age groups 16-25 and 25-40 years. The realist approach allows the opportunity for participant to disclose perspectives, opinions and experiences. The conduct of the workshops will enable the generation of hypotheses on mechanisms by which care in the last year of life for people aged 16-40 years might be delivered to enable ‘good’ care. We shall use our programme theories to underpin our thinking in the workshops and in the analysis of the data they generate. We shall use the emerging hypotheses to populate in more depth our existing programme theories. The teacher-learner approach by which researchers and participants both contribute to discussions will enable the researchers to use their theories and knowledge from the interviews to interact with workshop participants as well as confirm or falsify theories.

References

Agnew, A., Manktelow, R., Taylor, B. and Jones, L. (2009) Bereavement needs assessment in specialist palliative care: a review of the literature. *Palliative Medicine*, vol. 24:1, pp. 46-59.

Charmaz, K. (2006) *Constructing Grounded Theory: A Practical Guide Through Qualitative Analysis*. Sage, London.

Ferlie E.B, Shortell S.M. (2001) Improving the quality of health care in the United Kingdom and the United States: a framework for change. *The Millbank Quarterly*, vol. 79:2, pp. 281-315.

When Cure Is Not Likely – Interview Topic Guides

We need to collect some background information about each participant, so we can describe who has taken part in the study.
What year were you born?
How would you describe your gender?
Do you recall when you were first diagnosed (date/month/year?)
At the moment are you employed (on sick leave) or in full time education?
How would you describe your ethnicity?

During the interview, we would like to talk about a number of issues relating to your medical care, how this may have affected your social life, how well you think information has been given to you or how this could be improved and how decisions have been made about your care;

BOLD = questions Plain text = prompts

Patient	Past (up to WCINL) **Can you tell me what has happened up to this point?**	Present (WCINL) **What is currently happening?**	Future (Post Interview) **Do you think about things that might happen in the future?**
Medical Management	When did you find out something serious was wrong? [diagnosis] When did this happen, where, who was there, how did you feel What treatment did you have [if prompted by patient] How did you find out/ know things were not going so well [prognosis]	How do you think things are going with your illness /symptoms/ cancer at the moment? **Are you receiving any treatments now? Are you on medications, if so what are they for?** **What/who is helpful about the care you are receiving? What/who is unhelpful??** **How do you feel in yourself now? [feelings about medical aspects/situation]**	If yes, what do you think about / how do you plan?? Has anything been discussed with you about potential future treatment plans/ options [this needs to be dependent on what they say about the present]
Social Management	**How did you tell others (family/friends/colleagues) what was happening and how did they react** (illness, treatment, prognosis) **How did you find telling other people about your situation? If it was difficult, which parts were difficult??**	**Thinking about your day to day life – can you do the things that you want to do? If not, what is the impact on your life** **Does your illness have effects on you financially**?	**Is there anything in particular you want to do or achieve? (day to day; immediate, longer term)** **Do you have any worries/fears**

	On a very practical level, what were the biggest things that changed with the illness – e.g. where you were living / work / finance etc. **Have you used the internet to find out information about your illness** (What has been helpful/unhelpful? (illness, treatment) **Do you use social media like Facebook and Twitter? How has your illness affected that??**	**How does your current health affect you your relationships? (sexuality/family/friends/others/dependence/emotional)** **If you have a bad day, who/where do you turn to for support (what do they give you/how do they help? Have you found anything/ anyone particularly supportive)** **How do you feel in yourself now? [emotional wellbeing]** **Have you found a way to make some sense of what's happening at the moment? What has been helpful/unhelpful (spiritual)**	What are your fears (do you worry about) for the future? (parents/children/siblings, increase symptoms, being less able to do things you want to do, decreasing independence, dying process, life after death) **Do you have any hopes, dreams aspirations?**
Communication/information giving	**What was helpful / unhelpful about the conversations you have had with health professionals? How could it have been done better??** (e.g. timings / communication / information)	**Can you tell me about the support you are currently receiving from health care professionals (Cancer CNS, Pall Care, Consultant, GP, District Nurse, Social Worker)** **Can you tell me about the support you are receiving from your family (emotional, practical, financial, care)**	**What have your clinical team said about your future?** **What have you asked your clinical team about your future?** **Who do you think you would contact if you if you have concerns about the future? Why that person?**
Decision making	**Who has been involved in the decisions about your illness / care**?	**Are there any key decisions you are facing at the moment?**	**Are there any key things you think you will need to make decisions about in**

	What input have you had in decisions made about you illness/care?	**(further/stopping treatment; ACP; breaking news to others e.g. small children)** Prompt: If so, is anyone helping you with this? Is there anyone you would like to help you? Are you able to talk to that person / people… if not why not	**the coming weeks and months?** Who do you think might be able to help you with this? Are you able to talk to those people? – if not why not, **Are there questions you want to ask but don't feel able to? What might help to make that easier??** Prompts: (Simple everyday things/activities; Preferred place of care; Preferred place of death; memory boxes, photos, Writing a will; Funeral planning)

Is there anything we haven't mentioned that you would like to talk about today?

Family

We need to collect some background information about each participant, so we can describe who has taken part in the study.

How old are you?
How would you describe your gender?
Do you recall when you were first diagnosed (date/month/year?
At the moment are you employed or in full time education?
How would you describe your ethnicity?

During the interview, we would like to talk about a number of issues relating to x's medical care, how this may have affected their and your social lives and interactions, how well you think information has been given to x or to you and whether this could be improved and how decisions have been made about x's care.

Family	Past **What has happened up to this point**	Present **What is currently happening**	Future **What do you expect to happen in the future…**
Medical Management	When did you find out something serious was wrong with x [diagnosis] When did this happen, where, who was there, how did you feel **How involved have you felt in their treatment and care?** **How would you rate x's experiences of the care (diagnosis, treatment,;** helpful **/ unhelpful –in terms of care delivery / care pathway?** **Were you/other family members offered any support at this time by the clinical team (explain illness, treatment)** What was offered, did this differ by family members what was helpful/unhelpful	What are your views towards the current care that x is receiving How do you feel x is coping at the moment **How do you feel at the moment?** Do you feel supported Is there anyone who is supporting you? Do you have anywhere to go or anyone to talk to if you feel low? **How are the rest of your family coping (explore individual members)** **Is there anything more or different that could be done to help or support you or other family members?**	**Has anyone offered support to think about the future? If yes – was it helpful / unhelpful? If no – would you want some support ? who from? What should it look like?? What would you like?** What do you think might be available to help you and the patient in the future? (Pall Care, CNS, GP) What worries or hopes do you have about this?

	[if prompted by family member] How did you find out/ know things were not going so well [prognosis]		
Social Management	**What was the impact of x’s illness on you/your family’s day to day life (changes in relationships; priorities; carry on as ‘normal’)** In what ways has x’s illness changed your family (closer, distant) **Who did you tell that x had cancer; how did they react; how did this make you feel?**	**What is the impact of x’s illness on day to day life (financial, emotional)** How do you think this has changed your relationship with a) x b) other family members Have you found a way to make some sense of what’s happening at the moment? How has this been? **Is there anything particular that sustains you when things are not going so well? (spiritual)**	**What are your fears/ hopes (do you worry about) for the future?** What support do you think you will need in the future (emotional, financial, care) What support do you think other family members may need
Communication/information giving	**What information were you given after x’s diagnosis; who gave you this information; what did they say; what this information sufficient?** Do you think they were the best person to tell you this, if not, who would have been better How was information communicated between a) you and x; and b) between the different members of the family What was helpful/ challenging **Were there any things that you**	**What do you know about x’s current situation** **What else would you like to know about their current situation; where/who would you go to find this out?** **Who do you turn to for support? Is it adequate, how could it be improved?? What should change??** Who do you share how you are feeling with (family, friends, counsellor); what do they provide	**Has x or their clinical team discussed with you about what might happen in the next weeks and months?** **Have you been offered any support for now or in the future? Is it adequate, how could it be improved? What should change?**

	couldn't talk about to some people; any people you could talk openly too?		
Decision making	**How involved were you in any decisions that x had to make about their treatment** Who decided on your level of involvement Would you have liked greater or less involvement?	**How involved are you in decisions that x has to think about or make (what are these decisions; what involvement have you had)**	**Have you thought about what will happen in the future?** Have you discussed the future with x; what have you talked about (Preferred place of care; Preferred place of death; memory boxes, photos, Writing a will; Funeral planning)

Is there anything we haven't mentioned that you would like to talk about today?

Heath Care Professional

We need to collect some information about each participant.
How old are you?
How would you describe your gender?
Could you tell me what your current job title is please?
How long have you been in this position?
How long have you worked in palliative care?
How long have you worked with the young adult population?
Have you completed any further training for working with young adults with cancer?

During the interview, we would like to discuss the past, present and future dimensions of care for x, and then talk about your wider experience of working with this patient population. With these topics in mind;

With reference to the patient:

- **Past:** How long have you known the patient; What has happened up to this point? (Diagnosis, treatment)
- **Present:** What does the patient/ family understand about what is happening; at what points has communication of significant issues around what is happening taken place, can you tell me more about these. What discussions about this patient have taken place across the MDT
- **Future:** What do you expect to happen in the future? (Prognosis, towards end of life); what has been put in place for the patient – ACP? What support do you think the family will need – has anything been put in place? What is difficult for the patient?
- What do you think would improve the pathway of care for this patient?? How should/could it be done differently?

Wider experiences of working with young adult population

- What are the most important aspects of care for this patient group? (holistic; pain/symptoms; family issues; socio-demographic; financial, Peers, life tasks, support, advocacy, listening, empathy compassion , being there)
- Beyond providing medical care, what other needs do this patient group often have and how do you identify and address these e.g. social, family, educational, financial needs do you think that your system is robust enough, or is there the potential for issues areas to be missed / forgotten?
- At what stage do you know 'when cure is not likely'; can you give me examples of breaking this news to a patient/their family - where it went well and an example of where it was more challenging. What can make these scenarios more difficult
- How do palliative treatments e.g. chemotherapy affect patients' desired end of life outcomes (Ref, USA evidence chemo in last months of life associated with CPR ventilation and dying in ICU, Wright et al, 2014)
- How do you negotiate patient/family relationships (e.g. family dynamics)
- What do you enjoy about working with this population?
- What do you find more difficult about working with this population

- How does it affect you when a relatively young cancer patient dies (emotional impact; coping mechanisms; specific patients e.g. those with young children)

- Have any personal or professional experiences affected your practice (e.g. using experiences to improve care; managing emotions; maintain a professional distance)

- Do you have clinical supervision/support available; do you use this or other sources of support? How else do you relax and gain perspective

- Any issues you are aware of generally for professionals, teams, units, places of care, caring for young adults with cancer who may die

- What could be put in place to improve end of life care for this specific patient group?

Are there any other aspects of care for this population that we have not discussed and you feel are important?

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Understanding care when cure is not likely for young adults who face cancer: a realist analysis of data from patients, families and healthcare professionals.

Supplementary file 3 - Our approach to a realist logic of analysis.

Data analysis involved the use of a realist logic analysis with the goal of using the collected data (e.g. interviews) to confirm, refute or refine (test) aspects of our preliminary programme theory. Analysis required interpretation and judgement of data. Data coding was be deductive (informed by our preliminary programme theory), inductive (came from the data within data sources) and retroductive (where inferences are made based on interpretations of the data within data sources about underlying causal processes – i.e. mechanisms). We had used the data collected to develop themes. We then use a different analytical lens (a realist logic of analysis) to reanalyse the data we had used to develop our themes. More specifically, we used a series of questions to help us analyse the data, as set out below:

Relevance:
- Are sections of text within the collected data that are relevant to programme theory development or testing?

Interpretation of meaning:
- If the section of text is relevant, do its contents provide data that may be interpreted as functioning as context, mechanism or outcome?

Interpretations and judgements about Context-Mechanism-Outcome-Configurations:
- For the data that has been interpreted as functioning as context, mechanism or outcome, which Context-Mechanism-Outcome-Configuration (CMOC) (partial or complete) does it belong to?
- Are there further data to inform this particular CMOCs contained within this source or other sources? If so, which other sources?
- How does this particular CMOC relate to other CMOCs that have already been developed?

Interpretations and judgements about programme theory:
- How does this particular (full or partial) CMOC relate to the programme theory?
- Within this same source are there data which informs how the CMOC relates to the programme theory? If not, are there data in other sources? Which ones?
- In light of this particular CMOC and any supporting data, does the programme theory need to be changed?

Data to inform our interpretation of the relationships between contexts, mechanisms and outcomes were sought not just within the same data source, but across sources (e.g. mechanisms inferred from one source could help explain the way contexts influenced outcomes in a different source). Synthesising data from different sources is often necessary to compile CMOCs, since not all parts of the configurations will always be articulated in the same source.

Within the analytic process set out above, we used interpretive cross-case comparison to understand and explain how and why observed outcomes have occurred, for example, by comparing contexts where young adults had a 'better' end-of-life care experience with those where this was not to case. This enabled us to understand how context had influenced outcomes and why. When working through the questions set out, where appropriate we used the following forms of reasoning to make sense of the data:

- Juxtaposition of data: for example, where data about context in one source enabled insights into data about outcomes in another source.
- Reconciling of data: where data differed in apparently similar circumstances, further investigation was appropriate in order to find explanations for why these differences had occurred.
- Adjudication of data: on the basis of the plausibility of what was reported.
- Consolidation of data: where outcomes differed in particular contexts, explanations were constructed of how and why these outcomes occur differently.

During the evaluation, we moved iteratively between the analysis of particular examples, refinement of programme theory, and further data collection to test particular theories.

Reporting checklist for qualitative study.

Based on the SRQR guidelines.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the SRQR reporting guidelines, and cite them as:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. Standards for reporting qualitative research: a synthesis of recommendations. Acad Med. 2014;89(9):1245-1251.

		Reporting Item	Page Number
	#1	Concise description of the nature and topic of the study identifying the study as qualitative or indicating the approach (e.g. ethnography, grounded theory) or data collection methods (e.g. interview, focus group) is recommended	6-7
	#2	Summary of the key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results and conclusions	4
Problem formulation	#3	Description and signifcance of the problem / phenomenon studied: review of relevant theory and empirical work; problem statement	6-7
Purpose or research question	#4	Purpose of the study and specific objectives or questions	6-7
Qualitative approach and research paradigm	#5	Qualitative approach (e.g. ethnography, grounded theory, case study, phenomenolgy, narrative research) and guiding theory if	6-7

		appropriate; identifying the research paradigm (e.g. postpositivist, constructivist / interpretivist) is also recommended; rationale. The rationale should briefly discuss the justification for choosing that theory, approach, method or technique rather than other options available; the assumptions and limitations implicit in those choices and how those choices influence study conclusions and transferability. As appropriate the rationale for several items might be discussed together.	
Researcher characteristics and reflexivity	#6	Researchers' characteristics that may influence the research, including personal attributes, qualifications / experience, relationship with participants, assumptions and / or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results and / or transferability	n/a – semi-structured interviews and workshops
Context	#7	Setting / site and salient contextual factors; rationale	7
Sampling strategy	#8	How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g. sampling saturation); rationale	7-8
Ethical issues pertaining to human subjects	#9	Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	10
Data collection methods	#10	Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources / methods, and modification of procedures in response to evolving study findings; rationale	7-10
Data collection	#11	Description of instruments (e.g. interview guides,	7-10,

instruments and technologies		questionnaires) and devices (e.g. audio recorders) used for data collection; if / how the instruments(s) changed over the course of the study	supplementary upload
Units of study	#12	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	10
Data processing	#13	Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymisation / deidentification of excerpts	9
Data analysis	#14	Process by which inferences, themes, etc. were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale	9
Techniques to enhance trustworthiness	#15	Techniques to enhance trustworthiness and credibility of data analysis (e.g. member checking, audit trail, triangulation); rationale	9-10
Syntheses and interpretation	#16	Main findings (e.g. interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	12-16
Links to empirical data	#17	Evidence (e.g. quotes, field notes, text excerpts, photographs) to substantiate analytic findings	12-16
Intergration with prior work, implications, transferability and contribution(s) to the field	#18	Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application / generalizability; identification of unique contributions(s) to scholarship in a discipline or field	21-25
Limitations	#19	Trustworthiness and limitations of findings	25

Conflicts of interest	[#20](#)	Potential sources of influence of perceived influence on study conduct and conclusions; how these were managed	25
Funding	[#21](#)	Sources of funding and other support; role of funders in data collection, interpretation and reporting	Cover sheet and upload

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