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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

Pearce, SM

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#	Author	Affiliation	Email address
1*	Charlotte Kenten	University College Hospital, Cancer Clinical Trial Unit, London, NW1 2PG, UK	<a href="mailto:charlotte.kenten@nhs.net">charlotte.kenten@nhs.net</a>
2*	Nothando Ngwenya	University College Hospital, Cancer Clinical Trial Unit, London, NW1 2PG, UK	<a href="mailto:nothando.ngwenya@cantab.net">nothando.ngwenya@cantab.net</a>
3	Faith Gibson	Great Ormond Street Hospital for Children NHS Foundation Trust and University of Surrey, Guildford	<a href="mailto:Faith.Gibson@gosh.nhs.uk">Faith.Gibson@gosh.nhs.uk</a>
4	Mary Flatley	St Joseph's Hospice, London	<a href="mailto:mary@glynnflatley.com">mary@glynnflatley.com</a>
5	Louise Jones	Marie Curie Palliative Care Research Department, Division of Psychiatry, UCL	<a href="mailto:caroline.jones@ucl.ac.uk">caroline.jones@ucl.ac.uk</a>
6	Susie Pearce	Plymouth University School of Nursing and Midwifery Faculty of Health Sciences Drakes Circus, Plymouth PL48AA	<a href="mailto:Susie.pearce@plymouth.ac.uk">Susie.pearce@plymouth.ac.uk</a>

#	Author	Affiliation	Email address
7	Geoff Wong	Nuffield Department of Primary Care Health Sciences, University of Oxford, Oxford, OX2 6GG, UK	<a href="mailto:grckwong@gmail.com">grckwong@gmail.com</a>
8	Kath M Black	St Gemma's Hospice, Leeds	<a href="mailto:Kath.black@nhs.net">Kath.black@nhs.net</a>
9	Sue Haig	Southampton General Hospital Tremona Road Southampton SO16 6YD, UK	<a href="mailto:Sue.Haig@uhs.nhs.uk">Sue.Haig@uhs.nhs.uk</a>
10	Rachael Hough	University College Hospital, Children and Young Peoples Cancer Service, London, NW1 2PG, UK	<a href="mailto:rachaelhough@nhs.net">rachaelhough@nhs.net</a>
11	Adam Hurlow	Leeds Teaching Hospitals NHS Trust, Leeds	<a href="mailto:a.hurlow@nhs.net">a.hurlow@nhs.net</a>
12	L. Caroline Stirling	Central and North West London NHS Trust, Camden, Islington ELiPSe and UCLH & HCA Palliative Care Service, NW1 2PU	<a href="mailto:caroline.stirling@nhs.net">caroline.stirling@nhs.net</a>
13	Rachel M Taylor	University College Hospital, Cancer Clinical Trial Unit, London, NW1 2PG, UK	<a href="mailto:rtaylor13@nhs.net">rtaylor13@nhs.net</a>
14	Adrian Tookman	Marie Curie Hospice, Hampstead, London	<a href="mailto:Adrian.Tookman@mariecurie.org.uk">Adrian.Tookman@mariecurie.org.uk</a>
15	Jeremy Whelan	University College Hospital, Dept. of Oncology, London, NW1 2PG, UK	<a href="mailto:Jeremy.whelan@nhs.net">Jeremy.whelan@nhs.net</a>
* These authors contributed equally			

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**Correspondence:** [Jeremy.whelan@nhs.net](mailto:Jeremy.whelan@nhs.net)

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2

3

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4

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13 Armes and Mr John Reeve for their advice and oversight of the study as members of the  
14 External Steering Committee.

15

#### 16 **Author Contributions**

17 CK, NN, FG, MF, LJ, SP, GW, KMB, SH, RH, AH, LCS, RMT, AT, JW were all involved in  
18 study design, review of results and review of this manuscript. Interviews and analysis were  
19 conducted by CK and NN: aided by data analysis meetings with LJ, SP, FG, and GW.

20 Workshops were attended by CK, NN, MF, LCS, RH, FG, JW: KB and ABH attended 1  
21 workshop and led another. Participant recruitment was assisted by MF, KB, SH, AH, LCS,  
22 JW.

23

#### 24 **Data sharing statement**

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

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1 **Abstract**

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

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

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

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

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

24 Conclusions: Current care towards end of life for young adults with cancer and their families  
25 does not meet needs and expectations. We identified challenges specific to those aged 16-  
26 40 years. The burden that care delivery imposes on healthcare professionals must be  
27 recognised. These findings can inform recommendations for measures to be incorporated  
28 into services.

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**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.

## 1 **Background**

2 Cancer in young adults under 40 years is notable because it comprises a wide range of  
3 malignancies, has specific challenges to improving both length and quality of life, but is  
4 relatively uncommon.(1) One quarter of all deaths in the United Kingdom in people aged 16-  
5 40 years are from cancer.(2) In Europe there are over 27,000 deaths per year in this age  
6 group.(3) Despite increasing empirical evidence of the specific needs of young adults in  
7 specialist cancer care, there is little evidence about their experiences towards the end-of-life.  
8 (4-6)

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

15  
16 Concerns about improving end-of-life care are not confined to young adults. A recent  
17 interdisciplinary report published by the Royal College of Physicians in the UK summarises  
18 the concerns expressed by professionals, patients, families and other stakeholders such as  
19 charities. This report suggests that much more can be done to overcome barriers and myths  
20 that have been long-identified. The value of the perspective brought by patients and families  
21 is highlighted as a means to bring timeliness and honesty to discussions about dying whilst  
22 at the same time accounting for and respecting specific circumstances set by factors such as  
23 underlying disease, faith and as addressed here, age. (11)

24  
25 Boundaries between curative and palliative cancer treatments are often blurred as decisions  
26 may be influenced by cancer type, age and family circumstances as well as the experience  
27 and skills of healthcare professionals (HCPs). Avoidance and delaying of discussions about  
28 planning for care as health deteriorates and end-of-life decisions are common, often affecting  
29 the quality of care.(12) Professionals consistently acknowledge the challenges of managing  
30 end-of-life care for younger people, which may have commonalities with and, importantly,  
31 differences from those people with cancer at older ages as death approaches. (9, 10, 13)

32  
33 This work considers both the problem of limited data available in the literature and the  
34 desirability of understanding the experience of facing a poor prognosis at a young age from  
35 multiple perspectives. We wished to understand what were the core components in the  
36 pathways of care in the last year of life for people with cancer aged 16-40 years; whether  
37 there were any differences between the experiences of people with cancer from the age

1 ranges 16-24 and 25-40 years; how young adults and their families can be supported in the  
2 last year of life to achieve their preferences for care; and what challenges exist for health  
3 and social care professionals providing care.

4

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

13

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

33

34 Our preliminary programme theory was:

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



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

3  
4 We used this theory to develop topics for use in semi-structured interviews with young adults  
5 with cancer, family members and HCPs, and to underpin scenarios used in workshop  
6 discussions with HCPs and bereaved family members. That is, our preliminary programme  
7 theory sets out our initial hypotheses of the differences we thought were likely to set apart  
8 the end-of-life care experiences and preferences for younger people. Our interviews were  
9 thus developed by the project team in such a way as to be able to gather data that would  
10 enable us to confirm, refute or refine aspects of our programme theory. For example,  
11 because we hypothesised that a sense of control might influence end-of-life care  
12 experiences, we deliberately developed interview questions that asked about this issue. An  
13 important point about our initial programme theory is that it was refined as the evaluation  
14 progressed based on data gathered. As such, our expectation was that our preliminary  
15 programme theory would need to be refined to have adequate explanatory value.

16  
17 In this paper, we describe data arising from these interviews and workshops. We used our  
18 data analysis to further explore and develop realist causal explanations that may explain  
19 parts of our preliminary programme theory. As is expected in realist evaluations, as the  
20 evaluation progressed, we developed a revised programme theory that can be used to  
21 underpin recommendations for policy and practice and inform future research.

22  
23 **Methods**  
24 A multi-method realist study was undertaken (Figure 1). A realist evaluation approach was  
25 used as we wanted to explain and understand contextual influences on the experiences of  
26 and preferences for care towards the end-of-life for those with cancer aged 16-24 and 25-40.  
27 Here we report on Phases ii-iv, using RAMESES standards for reporting realist evaluations.  
28 (17) Phase v will be reported separately.

29  
30 **Recruitment and participants**  
31 We aimed to recruit a purposive sample of young people aged 16-40 with cancer, in two  
32 cohorts with an expected prognosis of less than one year, across four cancer groups:  
33 carcinomas; leukaemia and lymphoma; bone and soft tissue sarcoma; and central nervous  
34 system (CNS) tumours, which account for more than three-quarters of cancers occurring in  
35 this age group. Estimation of prognosis was made at each site by clinicians involved in  
36 screening and identifying people with cancer for the study. They used clinical records, their  
37 own clinical knowledge of disease progression and liaised with other members of the clinical

1 team to confirm, at the time of approach, that the prognosis for each individual was likely to  
2 be less than one year. In cohort 1 we planned to recruit a maximum of 15 participants aged  
3 16-24 years, including a minimum of three participants from each of the cancer groups, to be  
4 interviewed at two time points; recruitment began via a national cohort study investigating  
5 whether specialist cancer services add value ([www.brightlightstudy.com](http://www.brightlightstudy.com)) and was later  
6 extended, due to poor recruitment, to include five principal treatment centres and a hospice  
7 for young adults. Cohort 2 was recruited from three specialist cancer services and three  
8 hospices in England and consisted of a maximum sample of 30 participants between the  
9 ages of 16-40. All cohort 2 participants were invited to nominate a family member and HCP  
10 involved in their care for interview. The first-hand clinical experience of many in the project  
11 team aided the development of the study. Knowing that this is an under-researched  
12 population within the context of the study and drawing on professional experience to guide  
13 data collection, analysis and interpretation was essential. Further details are available in the  
14 protocols (supplementary files 1 and 2).

15

16 Data collection

17 Semi-structured interviews

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

27

28 Workshops

29 We held workshops in London, Southampton and Leeds. The workshops involved the  
30 participants sitting as one group. One clinical member of the team acted as the facilitator for  
31 the HCP workshops and two clinical members of the team were co-facilitators for the  
32 bereaved relative workshops. The co-facilitation meant that if someone from the group  
33 needed to leave or have a break from the discussion they could be supported by one of the  
34 co-facilitators whilst the workshop was able to continue. At the start of the workshop the  
35 facilitator introduced the study, outlined the workshop and informed consent obtained. The  
36 HCP workshops focused around the scenarios and the perspectives of different professional  
37 roles. The bereaved relative groups were guided by one of the facilitators with the

1 participants sharing narratives around their experiences with other participants either  
 2 supporting the narrative or outlining how their experience differed.

3

4 1. Healthcare professionals

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

11

12 2. Bereaved relatives

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

18

19 Table 1. Scenarios used in health care professionals' workshop

<b>Scenario 1: 16-40 year old patient with haematological malignancy</b>	<b>Scenario 2: 25-40 year old patient with oncological malignancy</b>
<p>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.</p> <p><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.</p> <p><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.</p> <p><u>November 2014</u>                      Inpatient. Deteriorating – wants to be at home. Unable to do stairs therefore need to make adaptations.</p> <ul style="list-style-type: none"> <li>• Symptoms – shortness of breath,</li> </ul>	<p>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.</p> <p><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.</p> <p><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 young child and partner as she deteriorates.</p> <p><u>September 2014</u>                      Helen has lost weight and is aware that she is getting weaker and has difficulty picking</p>

<p>cough and fatigue.</p> <ul style="list-style-type: none"> <li>• Care – family keen to do.</li> <li>• Discharge home with Community Palliative Care Team input.</li> <li>• Contact with charities - Willow Foundation, CLIC Sargent</li> </ul> <p><u>December 2014</u> Increased fatigue. Treated with radiotherapy to chest. Cough and fatigue.</p> <p><u>January 2015</u> Further deterioration. Bed bound. Home oxygen. Anticipatory medications.</p>	<p>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.</p> <p><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'</p> <p>Advanced care planning with clinical nurse specialist causes tension as parents do not wish Helen to be 'not for resuscitation'</p> <p><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.</p>
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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

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

5  
6 Re-analysis and re-interpretation of the themes to develop CMO configurations was  
7 undertaken by CK and NN aided by data analysis meetings with LJ, SP, FG, and GW. To  
8 assist the re-analysis and re-interpretation process, we attempted to develop CMO  
9 configurations that explained the outcomes in as many parts as possible of our  
10 preliminary programme theory; of the care pathways and experiences of people with  
11 cancer, family members and HCPs. For each of these mini programme theories we re-  
12 analysed the data that we drew on to develop each theme to build context-mechanisms-  
13 outcome (CMO) configurations - i.e. develop realist causal explanations of outcomes that  
14 occurred within different contexts (e.g. social rules and cultural systems). Workshop data  
15 were analysed in the same two-step manner and used to confirm, refute or refine the  
16 CMO configurations within the 'mini' programme theories.

#### 17 18 Ethical review

19 The study was approved by Central London Research Ethics Committee (Reference:  
20 13/LO/1098) and informed consent was sought from all participants at the time of  
21 participation.

#### 22 23 Patient and Public involvement

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

#### 33 34 **Results**

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

6 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	

7

8

9 The results are presented in three sections:

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

15

1 **Section 1 Thematic analysis**

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

4

5 **Loss of Control**

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

12

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

20 **Maintenance of Normal Life**

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

26

27 *I'm at probably the worst stage I've ever been with this illness, obviously because it's more*  
28 *advanced. Yet people are just saying, "You're looking great." And when I look in the mirror, I*  
29 *don't feel like I've got cancer. I don't feel like – obviously I do because I know that I do, but I*  
30 *don't feel any different to how I used to feel. Obviously yes you've got a few aches and pains*  
31 *and stuff, but you think like, when you hear someone's dying of cancer, you think that person*  
32 *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*  
33 *disconnect of like how – you know, like when you're feeling alright and you're going round*  
34 *doing stuff, and you're just doing stuff like everyone else, you just kind of forget. You go to*  
35 *work and you just have the same sort of, do the same things you were doing before you had*  
36 *cancer. You just forget, I forget sometimes (Cohort 2 – Patient 19)*

## 1 **Continuity of Care**

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

9  
10 *So we went into this initial meeting and [1st tumour CNS], who is the CNS, was there. And*  
11 *Dr [Consultant] was the one that kept us waiting. And it was said at that point, “[1st tumour*  
12 *CNS] will be your CNS, presumably key worker, throughout this process, she will be at every*  
13 *one of your appointments when you come to clinic.” And I was like, great, and he gave me*  
14 *her number and a pack and, you know, I felt quite supported by that. ... I understand not*  
15 *being able to the same nurse every time, that’s not possible, but like if you had a team that*  
16 *were allocated a certain number of patients – because they just, they don’t know you. And*  
17 *I’ve noticed that across the course of having another lot, you know, and I’ve really – I’ve kind*  
18 *of got to know a lot of them because I’ve been there, you know, over the course of a year.*  
19 *But, you know, it is at the beginning, it’s someone different every week. And they don’t know*  
20 *anything about you. And I went in expecting them to have read my notes, know what kind of*  
21 *cancer it was, know, you know, some of my background, and totally naively – they – and I*  
22 *think it’s unfair to them, they are there just to administer medication (Cohort 2 – Patient 29)*

## 23 **Professionals Need Support**

24 Professionals in either cancer or palliative care settings tended to have greater experience of  
25 caring for older adults. They had less experience providing end-of-life care to those aged 16-  
26 40 and fewer ‘tools’ or strategies to offer this younger population. Professionals found caring  
27 for young adults as they deteriorated both professionally and emotionally challenging and  
28 burdensome, as witness to young people prematurely reaching the end of their lives coupled  
29 with a weight of expectation to do more. The availability, accessibility and use of support for  
30 HCPs was variable and ranged from peer to professional support with a perception that  
31 experienced senior doctors were less likely to be in need. In contrast, nurses were perceived  
32 to be more likely to require and/or seek out support:

33  
34 *But there’s always been this sort of demarcation that when they come to the – come to,*  
35 *“They’re now incurable,” they go somewhere else. And that ‘somewhere else’ is always*  
36 *nebulous. ‘Someone else’ looks after them ‘somewhere else.’ Do you know what I mean? ...*



1 *“Oh they go over there now.” As I said earlier, the palliative team will look after them. And I*  
2 *don’t think any of us [Oncology CNS] have ever really gone to see what the palliative team*  
3 *do or see how much input they have. And is that a, is that a lack of professionalism or is that*  
4 *a survival mechanism for ourselves? And I have a feeling it’s the latter. I have a feeling that*  
5 *it’s very much a survival mechanism for ourselves because then we can just close that bit off*  
6 *and we can get on over here. And we’d like to know how they are, but we don’t have to be*  
7 *the one that tells them. (Cohort 2 – HCP 16)*

## 8 **Families Need Support**

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

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

16

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

## 24 **Language**

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

29 *I do remember him [Consultant] saying, I can’t really remember the conversation massively,*  
30 *but I do remember him keep saying, “Tumour, there’s a tumour.” And then I literally did have*  
31 *to say, “Hang on a minute, do you mean cancer?” and he said, “Yes, we’ve got to run more*

1 *tests and this, that and the other, but yes.” But that’s the only thing I remember really about*  
2 *it, if you know what I mean. (Cohort 2 – Patient 14)*

### 3 **Financial Concerns**

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

11  
12 *But you could do with somebody saying to you, in the first place, “You need somebody to*  
13 *help you to do this,” you know what I mean, you need somebody who can guide you through*  
14 *the system. And I think the same applied with [name]. He’d think, “Oh well I’ve just got to fill*  
15 *this form in and I’ve got...” but actually filling those forms in is a damned hard job. (Cohort 2*  
16 *– Family 23)*

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

### 21 **Section 2 Realist explanations of our themes presented in the form of Context-** 22 **Mechanisms-Outcome (CMO) configurations**

23  
24  
25 We reanalysed and re-interpreted our emergent themes using a realist logic of analysis. We  
26 attempted to identify mechanisms (generative causal processes) that are activated in the  
27 contexts we had found within the themes we uncovered. Our interview data were purely  
28 qualitative and so likely to be limited in the range of relevant data needed to build CMO  
29 configurations. To supplement these data, we deliberately drew on the extensive content  
30 expertise of the project team, workshops and where relevant, existing theories on needs of  
31 people living with cancer.

32 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
<p><b>CMO 1</b></p> <p>The diagnosis of cancer (context), changed the perception of control (mechanism) in young adults to cause distress, frustration and anger (outcomes).</p>	<p><i>... 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)</i></p>	<p>From the Loss of Control theme:</p> <p>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.</p>
<p><b>CMO 2</b></p> <p>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'.</p>	<p><i>"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]</i></p>	<p>From the Maintenance of Normal Life theme:</p> <p>Young adults wanted to live as normal a life for as long as possible.</p> <p>Young adults and their families adjusted to a new normality, to accommodate the changes their disease progression created.</p>
<p><b>CMO 3</b></p> <p>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</p>	<p><i>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</i></p>	<p>From the Continuity of Care theme:</p> <p>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:</p>

Care towards the end of life in young adults with cancer

<p>(mechanism) was less likely to occur.</p> <p>In contexts where continuity was provided (i.e. seeing the same HCP) trust developed (outcome) because of a sense of being known (mechanism).</p>	<p><i>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).</i></p>	
<p><b>CMO 4</b></p> <p>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).</p>	<p><i>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)</i></p> <p><i>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)</i></p>	<p>From the Professionals Need Support theme:</p> <p>The loss of a 'life partially lived' can be difficult for professionals, who feel a greater burden of sadness when young adults die</p> <p>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:</p>
<p><b>CMO 5</b></p> <p>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</p>	<p><i>It is quite upsetting because, we actually felt abandoned, I felt abandoned.</i></p> <p><i>[Facilitator: By?]</i></p> <p><i>By just the whole system really. It was just, if you</i></p>	<p>From the Families Need Support theme:</p> <p>Families often provided informal care for young adults within the home. Whilst they wanted to support their child or partner, caring created a</p>

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<p>them feeling marginalised (mechanism) resulting in feelings such as abandonment and distress (outcomes).</p>	<p><i>didn't ask, you wouldn't know (Family workshop 2- bereaved husband)</i>  <i>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)</i></p>	<p>further burden. Families felt insufficiently supported in this role.</p>
<p><b>CMO 6</b>                  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).</p>	<p><i>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).</i></p>	<p>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:</p>
<p><b>CMO 7</b>                  Few young adults have thoughts about long term financial planning as they did not anticipate serious illness (context). This can lead to individuals and</p>	<p><i>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</i></p>	<p>From Financial Concerns theme:                  Financial concerns and insecurity are almost always expressed by young adults with cancer.</p>

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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 crappiest 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**

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### **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 whilst 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**

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

#### 16 **Giving young people the chance to have control and to feel normal**

17 We found, perhaps unsurprisingly, the pivotal role of HCPs in supporting young adults with  
18 cancer. However, we were able to identify that an important ‘block’ to the support provided  
19 comes from the emotional discomfort felt by HCPs when discussing aspects of care  
20 specifically with young adults – such as discussions about prognosis. This is important as a  
21 cancer diagnosis creates great uncertainty and the knowledge that there will not be a cure  
22 creates a dissonance between the life that was expected and the reality of a life that will be  
23 significantly shorter than expected. For emerging adults and early independent adults, as  
24 disease progresses, dissonance is also present as their independence is compromised with  
25 an increasing and unanticipated dependence on others. This can affect their ability to attend  
26 school, college or work as well as taking part in family or social activities or fulfilling caring  
27 duties for others e.g. looking after young children. Adaptation is a mechanism through which  
28 there is a recognition of what can no longer be achieved due to disease progression.(22) An  
29 adapted normality can be achieved together with a sense of control, allowing for realistic  
30 goal setting.(23) Advance care planning could facilitate this adaptation. However, few  
31 participants in our study reported having had conversations about their options or the care  
32 they wanted to receive. Some HCPs avoided such conversations because of the emotional  
33 burden to themselves, not wishing to challenge either hope or a young person’s possible  
34 denial about their situation. This might be an example of what Bell et al refer to ‘as social  
35 constraint’, i.e. words and actions that inhibit end-of-life discussions.(24) A further notable



1 finding from the data indicates that all parties appear to wait for another to raise the topic of  
2 end-of-life. The 'window of opportunity' (17) often fails to appear, thus in some cases the  
3 topic is avoided. This has the potential to delay adaptation and limit the time available for  
4 professional support, which could help young adults plan and make as much as possible of  
5 remaining time.(25) For those with dependents, particularly young children, delaying  
6 adaptation could impact on their roles as parents, delaying the opportunity to prepare and  
7 create memories for themselves and their families.(23) When end-of-life was addressed, this  
8 tended to be when health had deteriorated, and that window of opportunity, albeit late,  
9 facilitated opportunities to discuss the future, end-of-life care and to make plans.

## 10 **Families and carers matter even more**

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

## 31 **The burdens for healthcare professionals**

32 Professionals reported difficulty addressing the needs of both the person with cancer and  
33 their family as often they had different expectations. Professionals were aware that providing  
34 bereavement support to a family was difficult if they had not built a relationship with them in

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

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

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

3

#### 4 **Strengths, limitations and future research directions**

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

#### 25 **Conclusion**

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

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1

## 2 **References**

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- 4 1. Bleyer A, Ferrari A, Whelan J, Barr RD. Global assessment of cancer incidence and  
5 survival in adolescents and young adults. *Pediatr Blood Cancer*. 2017;64(9).
- 6 2. What are the top causes of death by age and gender? [Internet]. 2012 [cited 02  
7 August 2016]. Available from:  
8 [http://webarchive.nationalarchives.gov.uk/20160105221750/http://www.ons.gov.uk/ons/rel/vs/ob1/mortality-statistics--deaths-registered-in-england-and-wales--series-dr-/2012/sty-causes-](http://webarchive.nationalarchives.gov.uk/20160105221750/http://www.ons.gov.uk/ons/rel/vs/ob1/mortality-statistics--deaths-registered-in-england-and-wales--series-dr-/2012/sty-causes-of-death.html)  
9 [of-death.html](http://webarchive.nationalarchives.gov.uk/20160105221750/http://www.ons.gov.uk/ons/rel/vs/ob1/mortality-statistics--deaths-registered-in-england-and-wales--series-dr-/2012/sty-causes-of-death.html).
- 10 3. Globocan 2012: Estimated Cancer Incidence, Mortality and Prevalence Worldwide in  
11 2012 [Internet]. 2012 [cited 02 August 2017]. Available from:  
12 [http://globocan.iarc.fr/Pages/fact\\_sheets\\_population.aspx](http://globocan.iarc.fr/Pages/fact_sheets_population.aspx).
- 13 4. Rosenberg AR, Wolfe J. Palliative care for adolescents and young adults with  
14 cancer. *Clin Oncol Adolesc Young Adults*. 2013;2013(3):41-8.
- 15 5. Ngwenya N, Kenten C, Jones L, Gibson F, Pearce S, Flatley M, et al. Experiences  
16 and Preferences for End-of-Life Care for Young Adults with Cancer and Their Informal  
17 Carers: A Narrative Synthesis. *J Adolesc Young Adult Oncol*. 2017;6(2):200-12.
- 18 6. Bleyer A, Barr R, Ries L, Whelan J, Ferrari A, editors. *Cancer in Adolescents and*  
19 *Young Adults*. 2nd ed. Switzerland: Springer; 2017.
- 20 7. Grinyer A. *Palliative and End of Life Care for Children and Young People: Home,*  
21 *Hospice, Hospital*. Oxford: Wiley; 2012.
- 22 8. Grinyer A, Thomas C. The importance of place of death in young adults with cancer. .  
23 *Mortality*. 2004;9(9):114-31.
- 24 9. Kars MC, Grypdonck MH, de Bock LC, van Delden JJ. The parents' ability to attend  
25 to the "voice of their child" with incurable cancer during the palliative phase. *Health Psychol*.  
26 2015;34(4):446-52.
- 27 10. Kars MC, Grypdonck MH, de Korte-Verhoef MC, Kamps WA, Meijer-van den Bergh  
28 EM, Verkerk MA, et al. Parental experience at the end-of-life in children with cancer:  
29 'preservation' and 'letting go' in relation to loss. *Support Care Cancer*. 2011;19(1):27-35.
- 30 11. Royal College of Physicians Our Future health. Talking about dying: How to begin  
31 honest conversations about what lies ahead. London: Royal College of Physicians; 2018  
32 October 2018.
- 33 12. Baker JN, Hinds PS, Spunt SL, Barfield RC, Allen C, Powell BC, et al. Integration of  
34 palliative care practices into the ongoing care of children with cancer: individualized care  
35 planning and coordination. *Pediatr Clin North Am*. 2008;55(1):223-50, xii.
- 36 13. Grinyer A, Barbarachild Z. *Teenage and young adult palliative and end of life care*  
37 *service evaluation*. London; 2011.
- 38 14. Pawson R, Tilley N. *Realistic evaluation*. London: Sage Publications Ltd; 1997. 235 ;  
39 23 cm. p.
- 40 15. The Rameses II Project. "Theory" in realist evaluation 2017 [Available from:  
41 [http://ramesesproject.org/pgm-](http://ramesesproject.org/pgm-download_media.php?name=RAMESES_II_Theory_in_realist_evaluation.pdf)  
42 [download\\_media.php?name=RAMESES\\_II\\_Theory\\_in\\_realist\\_evaluation.pdf](http://ramesesproject.org/pgm-download_media.php?name=RAMESES_II_Theory_in_realist_evaluation.pdf).
- 43 16. The Rameses II Project. Developing Realist Programme Theories 2017 [Available  
44 from: [http://ramesesproject.org/pgm-](http://ramesesproject.org/pgm-download_media.php?name=RAMESES_II_Developing_realist_programme_theories.pdf)  
45 [download\\_media.php?name=RAMESES\\_II\\_Developing\\_realist\\_programme\\_theories.pdf](http://ramesesproject.org/pgm-download_media.php?name=RAMESES_II_Developing_realist_programme_theories.pdf).
- 46 17. Wong G, Westhorp G, Manzano A, Greenhalgh J, Jagosh J, Greenhalgh T.  
47 RAMESES II reporting standards for realist evaluations. *BMC Med*. 2016;14(1):96.
- 48 18. Richards L, Qualitative S, Research. *Using NVivo in qualitative research*. London:  
49 Sage; 1999. ix, 218 pages : illustrations ; 25 cm. + 1 CD-ROM (4 3/4 in.) p.
- 50

- 1 19. Charmaz K. Grounded Theory: objectivist and constructivist methods In: Denzin NK,  
2 Lincoln YS, editors. Handbook of qualitative research. California: Sage Publications Ltd;  
3 2000. p. 1065 ; 26 cm.
- 4 20. Beerbower E, Winters D, Kondrat D. Bio-psycho-social-spiritual needs of adolescents  
5 and young adults with life-threatening illnesses: Implications for social work practice. Soc  
6 Work Health Care. 2018;57(4):250-66.
- 7 21. Soanes L, Gibson F. Protecting an adult identity: A grounded theory of supportive  
8 care for young adults recently diagnosed with cancer. Int J Nurs Stud. 2018;81:40-8.
- 9 22. Livneh H. Psychosocial Adaptation to Cancer: The Role of Coping Strategies.  
10 Journal of Rehabilitation. 2000;66(6):40-50.
- 11 23. Nissim R, Rennie D, Fleming S, Hales S, Gagliese L, Rodin G. Goals set in the land  
12 of the living/dying: a longitudinal study of patients living with advanced cancer. Death Stud.  
13 2012;36(4):360-90.
- 14 24. Bell CJ, Zimet GD, Hinds PS, Broome ME, McDaniel AM, Mays RM, et al.  
15 Refinement of a Conceptual Model for Adolescent Readiness to Engage in End-of-Life  
16 Discussions. Cancer Nurs. 2018;41(2):E21-E39.
- 17 25. Llewellyn H, Neerkin J, Thorne L, Wilson E, Jones L, Sampson EL, et al. Social and  
18 structural conditions for the avoidance of advance care planning in neuro-oncology: a  
19 qualitative study. BMJ Open. 2018;8(1):e019057.
- 20 26. Knox MK, Hales S, Nissim R, Jung J, Lo C, Zimmermann C, et al. Lost and stranded:  
21 the experience of younger adults with advanced cancer. Support Care Cancer.  
22 2017;25(2):399-407.
- 23 27. Mohammed S, Swami N, Pope A, Rodin G, Hannon B, Nissim R, et al. "I didn't want  
24 to be in charge and yet I was": Bereaved caregivers' accounts of providing home care for  
25 family members with advanced cancer. Psychooncology. 2018;27(4):1229-36.
- 26 28. Bell CJ, Skiles J, Pradhan K, Champion VL. End-of-life experiences in adolescents  
27 dying with cancer. Support Care Cancer. 2010;18(7):827-35.
- 28 29. Wiener L, Weaver MS, Bell CJ, Sansom-Daly UM. Threading the cloak: palliative  
29 care education for care providers of adolescents and young adults with cancer. Clin Oncol  
30 Adolesc Young Adults. 2015;5:1-18.
- 31 30. Clark JK, Fasciano K. Young adult palliative care: challenges and opportunities. Am J  
32 Hosp Palliat Care. 2015;32(1):101-11.
- 33 31. Bradford NK, Greenslade R, Edwards RM, Orford R, Roach J, Henney R.  
34 Educational Needs of Health Professionals Caring for Adolescents and Young Adults with  
35 Cancer. J Adolesc Young Adult Oncol. 2018.
- 36 32. Mills J, Wand T, Fraser JA. Self-Care in Palliative Care Nursing and Medical  
37 Professionals: A Cross-Sectional Survey. J Palliat Med. 2017;20(6):625-30.
- 38 33. Mills J, Wand T, Fraser JA. Examining self-care, self-compassion and compassion  
39 for others: a cross-sectional survey of palliative care nurses and doctors. Int J Palliat Nurs.  
40 2018;24(1):4-11.
- 41 34. Odejide OO, Cronin AM, Earle CC, Tulskey JA, Abel GA. Why are patients with blood  
42 cancers more likely to die without hospice? Cancer. 2017;123(17):3377-84.
- 43 35. Saunders B, Sim J, Kingstone T, Baker S, Waterfield J, Bartlam B, et al. Saturation in  
44 qualitative research: exploring its conceptualization and operationalization. Qual Quant.  
45 2018;52(4):1893-907.

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**Legends for figures**

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

Figure 2. Analysis process

1 Manuscript ID bmjopen-2018-024397

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

4

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

6

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

16

17 Relevance:

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

20

21 Interpretation of meaning:

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

24

25 Interpretations and judgements about Context-Mechanism-Outcome-Configurations:

26 - For the data that has been interpreted as functioning as context, mechanism or outcome,  
27 which Context-Mechanism-Outcome-Configuration (CMOC) (partial or complete) does it  
28 belong to?

29 - Are there further data to inform this particular CMOCs contained within this source or other  
30 sources? If so, which other sources?

31 - How does this particular CMOC relate to other CMOCs that have already been developed?

32

33 Interpretations and judgements about programme theory:

34 - How does this particular (full or partial) CMOC relate to the programme theory?

35 - Within this same source are there data which informs how the CMOC relates to the  
36 programme theory? If not, are there data in other sources? Which ones?

37 - In light of this particular CMOC and any supporting data, does the programme theory need  
38 to be changed?

39

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

46

47 Within the analytic process set out above, we used interpretive cross-case comparison to  
48 understand and explain how and why observed outcomes have occurred, for example, by

1 comparing contexts where young adults had a 'better' end-of-life care experience with those  
2 where this was not to case. This enabled us to understand how context had influenced  
3 outcomes and why. When working through the questions set out, where appropriate we used  
4 the following forms of reasoning to make sense of the data:

- 5
- 6 - Juxtaposition of data: for example, where data about context in one source enabled  
7 insights into data about outcomes in another source.
  - 8 - Reconciling of data: where data differed in apparently similar circumstances, further  
9 investigation was appropriate in order to find explanations for why these differences had  
10 occurred.
  - 11 - Adjudication of data: on the basis of the plausibility of what was reported.
  - 12 - Consolidation of data: where outcomes differed in particular contexts, explanations were  
13 constructed of how and why these outcomes occur differently.

14

15 During the evaluation, we moved iteratively between the analysis of particular examples,  
16 refinement of programme theory, and further data collection to test particular theories.  
17



Figure 1

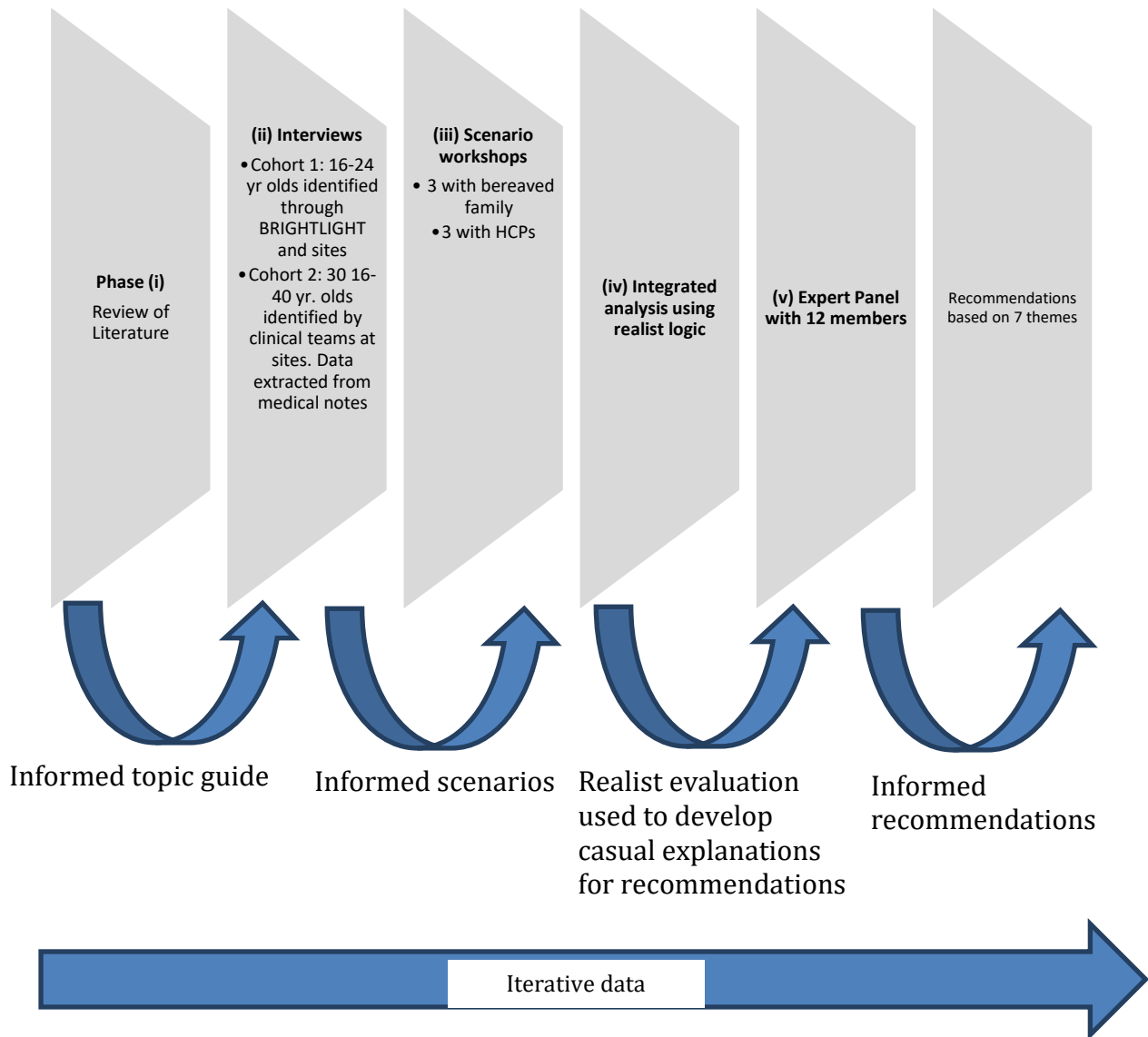
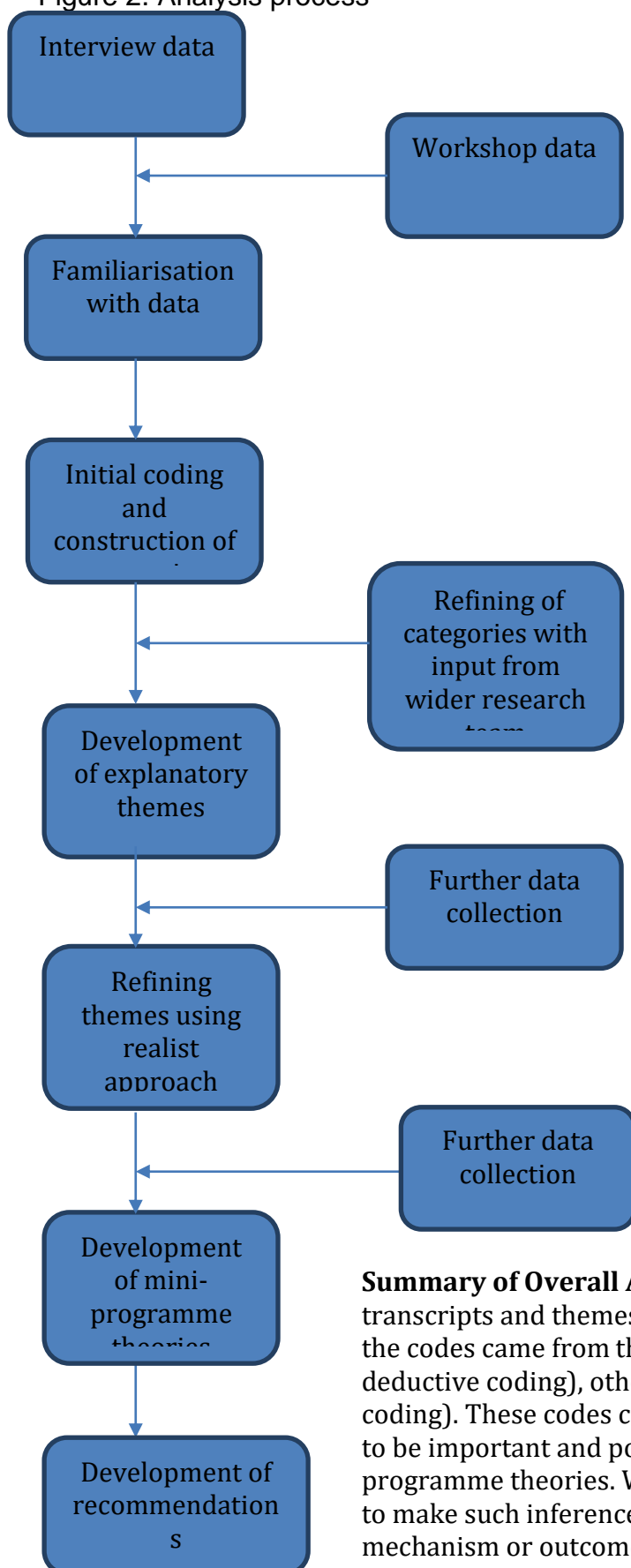


Figure 2. Analysis process



**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.