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Using qualitative and co-design methods to inform the development of an intervention to support and improve physical activity in childhood cancer survivors: a study protocol for BEing Active after ChildhOod caNcer (BEACON)

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3 **Using qualitative and co-design methods to inform the development of an intervention to support**
4 **and improve physical activity in childhood cancer survivors: a study protocol for BEing Active after**
5 **Childhood caNcer (BEACON)**
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ABSTRACT

Introduction:

Childhood cancer survivors (CCSs) treated with cardiotoxic cancer treatments are at increased risk of developing cardiometabolic complications. This risk is further exacerbated by poor health behaviours. In particular, CCSs are less active than non-cancer comparators. Existing interventions aiming to improve physical activity (PA) levels in CCSs are methodologically weak. The aim of this study is to rigorously and systematically develop an evidence- and theory-based intervention to promote, support, improve and sustain PA levels in CCSs, with the long-term goal of reducing CCSs' cardiovascular morbidity and mortality.

Methods and analysis:

The Being Active after Childhood Cancer (BEACON) study involves two workpackages at two NHS sites in England, UK. *Participants:* CCSs and their parents, and healthcare professionals (HCPs) involved in their care. *Methods:* Workpackage one (WP1) will use qualitative methods to explore and understand the barriers and facilitators to PA in CCSs. Two sets of semi-structured interviews will be conducted with (i) CCSs (aged 10 -24 years) and (ii) parents of CCSs. Workpackage two (WP2) will use co-design methods to bring together stakeholders (CCSs; their parents; HCPs; researchers) to develop a prototype intervention. Where possible, all data will be audio-recorded and transcribed. *Analysis:* Data from WP1 will be analysed using a thematic approach. Analysis of WP2 data will involve content analysis, and analysis of formative output and procedures.

Ethics and dissemination:

The study was approved by North East - Tyne & Wear South Research Ethics Committee (REC ref: 18/NE/0274). Research findings will be disseminated primarily via national and international conferences and publication in peer-reviewed journals. Patient and public involvement (PPI) will inform further dissemination activities.

ARTICLE SUMMARY

Strengths and limitations of this study

- The BEACON study will provide in-depth knowledge on the barriers and facilitators to PA in CCSs.
- The use of recognised frameworks of intervention development and principles from behavioural science to systematically develop an evidence- and theory-based health behaviour change intervention is a significant strength.
- Intervention development will actively engage stakeholders (CCSs, parents and healthcare professionals), ensuring that the resulting intervention is co-produced with those it aims to support, and maximizing likely acceptability and feasibility.
- The prototype intervention developed will be ready to be taken forward into production and testing.

INTRODUCTION

As a result of treatment advances, the population of childhood cancer survivors (CCSs) has rapidly grown. In the UK alone, there are more than 40,000 CCSs,¹ whilst across Europe there may be up to 500,000.² Two-thirds of CCSs may develop chronic health conditions by 15-25 years post-diagnosis.³ In particular, those treated with cardiotoxic therapies can experience persistent and cumulative damage to their cardiovascular, pulmonary and metabolic systems.⁴ Cardiovascular complications are a leading cause of morbidity among CCSs, and British CCSs have a 3.4-fold excess risk of cardiac death.⁵ Development of these chronic conditions impacts adversely not only on the survivors' physical health, but also on their psychological health and wellbeing and incurs costs for the healthcare system.

In the general population, it is well recognised that poor cardiovascular outcomes are strongly related to modifiable health behaviours, including a lack of physical activity (PA).⁶ Similarly, low levels of PA in even young CCSs (<18 years), have been linked to a worse cardiovascular risk profile,⁷ and CCSs are often less active than controls without a history of cancer.⁸ However, amongst long-term survivors of childhood lymphoma, a higher levels of vigorous PA is associated with a 50% lower risk of any cardiovascular event.⁹ Moreover, a Cochrane review indicated that physical exercise training programmes may improve physical fitness, body composition and cardiorespiratory fitness in childhood cancer patients and survivors.¹⁰ Considered together, this evidence provides a strong rationale for developing effective interventions to increase PA in CCSs.

Reviews have concluded that interventions to increase PA levels among CCSs are feasible and safe. However, studies are heterogeneous and most are methodologically limited.^{11,12} In addition, there is little evidence that interventions have been systematically developed using recognised frameworks of intervention development.¹³ Critically, while an understanding of factors which may promote or inhibit the target health behaviour (here, PA) is an essential first step in intervention design,¹⁴⁻¹⁷ most interventions appear to have been developed without having undertaken formative work to gain this understanding. Indeed, currently, little is known about determinants of PA behaviours in CCSs. Additionally, although the application of appropriate theory is recognised in behavioural science as an essential element of behaviour change interventions,¹⁴⁻¹⁷ most interventions appear to have no robust theoretical underpinnings.

Leading authorities advocate active stakeholder involvement in the design and development of novel health interventions,¹⁴⁻¹⁷; this is essential to understanding the perspectives and psychosocial context of users.¹⁸ However, most interventions for CCSs have been developed without the involvement of CCSs, raising concerns about the relevance and acceptability of the interventions to survivors. Moreover, although parents are key agents in their children's PA behaviour,^{19,20} there has been little attempt to understand either: (i) how the beliefs of parents of CCSs might influence their child's PA behaviours, or (ii) how parental support may be harnessed to encourage PA in CCSs.

A further consideration is wider implementation of interventions among CCSs. Those developed thus far were not designed to be deliverable within the context of the UK National Health Service (NHS). More generally, there is a lack of research exploring how support to modify health behaviours (including PA) among CCSs can be implemented effectively and feasibly in follow-up care. Involvement of HCPs and other relevant stakeholders in the development process would increase the likelihood that an intervention will be feasible, acceptable and implementable in the current healthcare pathway for CCSs; this does not appear to have been widely done.

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3 This project seeks to comprehensively investigate barriers and enablers to PA among CCSs – from
4 the CCS, parental and wider stakeholder perspective. Using the knowledge gained, and with the
5 support of key stakeholders, we will develop a person-centred evidence- and theory-based
6 prototype intervention aimed at promoting and supporting sustainable PA behaviour change in
7 CCSs.
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10 11 12 **AIMS AND OBJECTIVES**

13 14 **Aim**

15 The ultimate goal of the BEACON project is to develop an intervention which can reduce
16 cardiometabolic risk markers in the medium-term, and reduce cardiovascular morbidity and
17 mortality in the long-term, whilst also helping to prevent a deterioration in patient well-being and
18 health-related quality of life due to poor cardiovascular health.
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21 22 **Objectives**

23 The objectives of this phase of the BEACON project are to:

- 24 1) explore CCSs' experiences of, and participation in, PA behaviours.
- 25 2) identify and explore the barriers and enablers of PA behaviours in CCSs.
- 26 27 3) explore CCSs' and parents' experiences of receiving advice on PA or exercise, and perceived
28 need for this information.
- 29 30 4) actively engage key stakeholders (CCSs, their parents, HCPs) in a co-design process to
31 develop a prototype intervention.
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37 38 39 **METHODS AND ANALYSIS**

40 41 **Study design**

42 The study will be informed by intervention development approaches - notably the Medical Research
43 Council framework of intervention development¹⁵ and the Person-Based Approach¹⁸ to ensure that
44 the resulting intervention is systematically developed from the bottom-up and: 1) is theory- and
45 evidence-based;^{14 15 17 21-23} 2) prioritises and incorporates the views of the people who will use the
46 intervention;^{18 24} and 3) is likely to be implementable and scalable in the NHS.^{25 26} The current phase
47 of the project is focused on the intervention development. The planned research activities (which
48 form two sequential workpackages), and the other formative work previously undertaken by the
49 study team which will feed into intervention development, are summarized in Table 1 and Figure 1.
50 Workpackage 1 (WP1) will generate evidence on determinants of PA among CCSs. Workpackage 2
51 (WP2) will involve a co-design process to produce a prototype intervention.
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Participants

WP1 involves CCSs and parents/guardians of CCSs; eligibility criteria are shown in Table 2. Participants will be recruited via two clinical sites which are both specialist centres in childhood cancer treatment.

WP2 will recruit CCSs, parents/guardians of CCSs and HCPs. The same inclusion and exclusion criteria as for WP1 will apply for CCSs and parents/guardians. HCPs will be eligible if they are involved in the follow-up care of CCSs. Academics and researchers with relevant expertise will also be eligible.

Sampling

WP1: Generating evidence on determinants of PA among CCSs

Potential participants will be selected using purposive sampling with strata comprising of: age (for CCSs - current age of 10-15/16-24 years; for parents/guardians - age of child at diagnosis: ≤ 10 ; 11-18 years); clinical site); and cancer site (haematological malignancy/solid tumour/central nervous system tumour). Diversity in other characteristics (e.g. gender, treatment, time since diagnosis) will be sought to ensure sample heterogeneity and elicitation of a broad range of views and experiences.

Recruitment will continue until data saturation is reached in each interview set, defined as no new themes arising in the last three interviews.²⁷ We estimate that, interviews with 25-30 CCSs and 25-30 parents/guardians will provide adequate data. CCSs may participate without their parent/guardian taking part, and vice versa.

WP2: Co-design process

Up to 40 CCSs, parents/guardians of survivors, HCPs and academics/researchers will take part in the co-design process. At least 20 participants will be CCSs (due to the nature of the activities, participation is deemed only to be suitable for CCSs aged 16 and above) and 10 will be parents/guardians. As with the interviews, it will be important to seek diversity in the participants.

HCPs invited to take part in WP2 will include consultant oncologists and nurse specialists from paediatric and teenage and young adult services and other relevant HCPs (e.g. occupational therapists, physiotherapists, psychologists, cardiologists). The aim is that at least six HCPs will participate in the co-design process. Academics/researchers with expertise in the following fields will also participate: behavioural science, health psychology, PA and exercise science, healthcare technologies and human-centred design, and childhood cancer survivorship.

Identification, screening and recruitment of sample

Identification and screening of CCSs began in February 2019 with recruitment expected to end December 2020.

WP1: Generating evidence on determinants of PA among CCSs

Employing multiple recruitment strategies can guard against recruitment problems,²⁸ therefore, we will use up to three methods for recruiting participants. The primary method will require consultant oncologists/nurse specialists to screen attendance lists of forthcoming CCSs follow-up clinics at collaborating sites. At the clinic, eligible CCSs and their parents/guardians will be informed of the study by their child's oncologist/nurse specialist and asked if they would like to meet the researcher (MB). If so, the researcher (MB) will provide further details including the study information sheet and answer any questions. Potentially interested CCSs and parents/guardians will be asked whether the researcher can contact them in a few days to find out whether they would like to participate. Meeting the researcher at clinic will help potential participants feel more at ease and aid the establishment of rapport, which may be particularly important for younger patients.²⁹

Approvals are in place for variants of this process in the event that the researcher cannot attend the clinic, or the clinic is too busy for collaborating clinical colleagues to approach eligible CCSs and parents/guardians individually. These include (1) clinical colleagues recording details of those potentially interested of behalf of the researcher, who will follow-up by phone and (2) provision of study packs (containing a reply slip) to eligible CCSs and parents/guardians at clinic check-in with those interested returning the reply slip in a sealed envelope to the receptionist to forward to the researcher.

The second method will involve consultant oncologists/nurse specialists at the two sites screening patients in their care for eligibility and mailing a study information sheet; follow-up telephone calls by the clinical colleagues are permitted. Interested CCSs and parents/guardian may contact the researcher directly.

The third method, if required, will be identification of survivors via cancer registries in the study areas: the Northern Region Young Person's Malignant Disease Registry (NRYPM DR) and the Yorkshire Specialist Register of Cancer in Children and Young People (YSRCCYP).

Information sheets for CCSs are developmentally appropriate and designed for ages 10-12, 13-15 and ≥ 16 years. CCSs aged 10-15 years will also receive a copy of the Charter of Rights for Children and Young People in Research.³⁰ Parents of CCSs aged 10-15 years will receive an information sheet explaining the study their child has been invited to participate in.

WP2: Co-design process

WP1 participants will be asked if they wish to be notified of/invited to the co-design activities. New participants, without experience of the study, will also be recruited. Methods for recruiting CCSs and their parents/guardians will mirror those of WP1. In addition, we will also seek to recruit via social media, support groups and charities. Posts advertising the study will ask for interested individuals to contact the researcher who will assess eligibility and provide further information.

Eligible HCPs working at the collaborating sites will be invited to take part. We will also promote the study via social media, and through networks of the Childhood Cancer & Leukaemia Group, to encourage participation of HCPs from across UK. Academics and researchers experienced in relevant areas (also previously stated) will also be asked to participate.

Data collection

WP1: Generating evidence on determinants of PA among CCSs

Interviews with CCSs aged 10-15 years will take place face-to-face at the interviewee's and their parents preferred location (e.g. university/home). A parent/guardian may be present if they or the child wishes. These interviews are expected to last 30-60 minutes, but length will be determined by the child.

Interviews with CCSs aged ≥ 16 years, and those with parents/guardians, will take place by telephone, an end-to-end encrypted web app which enables secure audio/video calls (e.g. WhatsApp, Zoom), or face-to-face at a location of their choosing; providing choice on ways to participate can help maximise recruitment.²⁸ Experience suggests these interview will last 60-90 minutes³¹, but may be longer if the interviewee wishes.

Before the interview commences, the researcher will seek informed consent; for those aged 10-15 years, a parent/guardian will provide consent and the interviewee assent to ensure that the child feels involved in the decision about their participation.³² Participants (and parents on behalf of children aged 10-15 years) will be asked to complete a short demographic questionnaire. Time will be spent developing rapport and creating a secure, trusting environment,²⁸ particularly with younger children.²⁹

The interviews will be guided by a topic guide, which will be informed by the Theoretical Domains Framework (TDF), an integrative framework of behaviour change theories,^{21 22} and will cover: participant's views and attitudes towards their own/their child's PA; difficulties experienced with, and barriers to, PA; whether support/advice has been given regarding PA; and what helps or would help the survivor to be more active. Questions will be open and neutral. Topic guides will be used flexibly to allow interviewees to raise issues they consider important to the topic (PA); if this results in new areas, these will be explored in subsequent interviews to ensure sufficient depth is reached.

Interview content will be developmentally appropriate. Interviews with children aged 10-15 years will utilise cue cards and images to help engage and focus the participant and provide them with some control over the order of the questions.²⁹ With the interviewees' permission, interviews will be audio-recorded; if permission for recording is not granted, the researcher will take detailed notes.

WP2: Co-design process

We will follow the sequential and systematic co-design approach to integrate scientific evidence, expert knowledge and experience, and stakeholder involvement to design a prototype intervention.²⁴ The two phases of WP2 are described below.

Phase 1: Behavioural analysis

The researcher team will combine WP1 findings and other formative work previously conducted (e.g. a systematic review) (Figure 1), into a "theoretical model" of PA engagement among CCSs. They will identify which influences on PA are potentially modifiable to determine what needs to be done to change behaviours. The Behaviour Change Wheel will be used to map the TDF domains, and organise these into a working theoretical model of PA in CCSs.²³ For each identified factor the team will identify which intervention functions might be effective in changing PA behaviours. For each of

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2
3 the relevant intervention functions, associated behavioural change techniques will be identified (i.e.
4 the techniques that can be used to overcome barriers to, and enhance enablers of, engagement with
5 PA).³³
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8 *Phase 2: Co-design process*

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10 This process will involve a range of methods in order to engage and collaborate with stakeholders
11 flexibly. Modes of participation will include workshops (face-to-face or online), interviews (one-to-
12 one or small groups of 2/3 people; face-to-face or online); and online collaborative groups.
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15 Face-to-face workshops are expected to last 3-4 hours, while those online will be shorter (1-2 hours).
16 Interviews are likely to also last 1-2 hours. Video-conferencing is an acceptable method for
17 discussion with young people and an optimal alternative to face-to-face groups; it also enables
18 people from various geographical location to attend.³⁴
19

20 Online collaborative groups allow stakeholders to engage with the development process, and one
21 another, both in real time and asynchronously. Secure groups will be set up via WhatsApp, Facebook
22 or an Ideaboard.co.uk website developed specifically for the study. WhatsApp and Facebook are
23 widely used and familiar applications, and have successfully been used for co-design^{35 36}; Ideaboard
24 offers greater flexibility. Preferences of potential participants will inform the choice of platform. The
25 team will post content to the groups (e.g. videos, images, questions) and invite feedback.
26
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28 Following PPI input, CCSs will have the choice to participate in a survivor only or mixed (survivors
29 plus parents/HCPs) workshop/small group interview/collaborative group.
30

31 Using these methods, a range of activities (e.g. think aloud, mapping, brainstorming, storyboarding)
32 will be used to engage participants, provide ways for them to share, envision and develop their ideas
33 with others and to facilitate interaction. The specific activities will be dictated by the findings of
34 WP1, and the findings of any preceding workshops.
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36

37 Two steps in the co-design process are envisioned (Figure 1). In the first step, evidence statements
38 on PA among CCSs will be presented. Stakeholders' views on the relevance, importance and
39 effectiveness will be sought. Activities will be used to generate insights into what is needed to
40 improve PA levels in CCSs and novel intervention ideas which stakeholders think could be effective
41 and acceptable in improving PA in CCSs. Ideas will also be sought for how an intervention should be
42 designed, where and how it should be implemented, and the relevant components. Mapping
43 activities will enable organisation and visualisation of resulting intervention ideas and their key
44 components. The research team will analyse information collected to develop 'intervention
45 principles', ensuring that the evidence and theory, which is central to the success of the
46 intervention, remains intact.³⁷
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48

49 During the second stage activities will focus on gaining user feedback on intervention principles.
50 Content and mode of delivery will be further developed and refined. Intervention tailoring will also
51 be considered. Participants will identify and discuss potential challenges around acceptability,
52 usability and feasibility from different perspectives (e.g. CCSs, parents, HCPs, commissioners, service
53 providers). Based on participants discussion and decisions, designers/creative facilitators will begin
54 to sketch paper-based 'mock-ups' of the intervention.
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57 Outputs will be critically evaluated and translated into a design brief which details the aims of the
58 intervention, the design features it will include, and how these will be operationalised, taking care
59 will to ensure alignment with evidence and theory. A logic model will be developed,¹⁴ providing a
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graphical/textual representation of how the intervention is intended to work, linking outcomes with processes, the underlying theoretical assumptions and active ingredients (or the behaviour change methods and techniques that will be used to target the identified processes/mechanism associated with behaviour and behaviour change).^{33 38} The outcome will be a mock-up of the prototype which will represent the main features of the intervention. This prototype will be ready to take into production and undergo refinement and optimisation before going forward into further testing in a future study.

Prior to participation in co-design activities, informed consent will be sought and a ground rules for communication and engagement established. The researchers will ensure an atmosphere which is welcoming and non-judgemental and will be clear that all participants are treated as equals whether they are young people, parents, researchers or HCPs.

Where possible, co-design activities will be audio or video recorded and transcribed. Other data collected will include written data/notes, mapping activities and sketches resulting from the various activities (e.g. group work, brainstorm) and written comments generated by the online collaborative groups.

Data analysis plan

WP1: Generating evidence on determinants of PA among CCSs

Interview recordings will be transcribed verbatim. Analysis will occur in parallel with data collection to ensure that any new issues raised are explored in subsequent interviews. Interviews from CCSs and parents/guardians will be analysed separately. To identify views and experiences of, and barriers and facilitators to, PA in CCSs an inductive thematic analysis will be conducted.³⁹ Two team members will code data from preliminary interviews and discuss and agree the emerging codes and potential themes. Codes relating to the barriers and facilitators to PA will be mapped onto the TDF.^{21 22} These codes will then be applied by the researcher to remaining interviews, incorporating any new codes and themes as they are identified. For analytical rigour, the classification of belief statements to the TDF domains will be discussed and agreed within the team. Coding and analysis will be facilitated by QSR International's NVivo software (Version 12, 2018).

WP2: Co-design activities

Analysis of co-design activities will be focused specifically on the aims of each activity, pragmatic and expeditious so findings can be fed into subsequent stages.³⁷ Qualitative content analysis will be performed using QSR International's NVivo software (Version 12, 2018), supplemented by other forms of analysis as required.

Ethics

A favourable opinion has been granted from the North East - Tyne & Wear South Research Ethics Committee (REC ref: 18/NE/0274).

Informed consent will be sought prior to participation. Participants will be informed that participation is entirely voluntary, and they may withdraw at any point, without giving a reason and without negative consequences. They will be asked for their agreement to audio/video record

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3 (where relevant) and informed that recordings are confidential, and transcriptions of audio-
4 recordings will be anonymised. Ethical considerations relating to the interviewing of children (aged
5 under 16 years) are described above. Interview participants will be offered payment of any travel
6 expenses and a high-street shopping voucher. Participants in co-design activities will receive
7 payment in line with INVOLVE guidance.⁴⁰
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10 11 12 **Patient and public involvement and project steering group**

13 Feedback on the study concept and methods were gained from Young Person's Group – North
14 England (YPAG-NE), an established adult cancer survivor group (Perspectives) and two European
15 organisations for childhood cancer survivors. YPAG-NE and two adult CCSs provided comments on
16 patient information and interview topic guides/cue cards. Patient representatives sit on the project
17 steering group, and will be involved in data interpretation, co-design activities, and advising on
18 dissemination.
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22 23 **Dissemination**

24 Findings will be disseminated via our study website (<https://research.ncl.ac.uk/beingactive/>),
25 conferences and journal publication. A summary of research findings will be available for
26 participants. PPI will inform further dissemination activities (e.g. via patient organisations, social
27 media), appropriate formats (e.g. infographics, video) and content to ensure lay summaries are
28 understandable and engaging to survivors.
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32 33 **DISCUSSION**

34 A high proportion of adolescents and young adults do not meet recommended levels of PA,⁴¹ and
35 there are already many publicly available programmes and interventions to encourage PA, including
36 government/health service initiatives and apps.⁴²⁻⁴⁴ This raises the question of whether a specific PA
37 intervention is needed for CCS. While there is considerable overlap between determinants of PA in
38 CCS and young people without cancer, many influences are likely specific to CCS (e.g. cancer-related
39 fatigue, frustration about impact of cancer).¹³ There are also concerns about the quality and likely
40 effectiveness of many of the publicly-available PA programmes.⁴⁴ In addition, cancer survivors may
41 question the relevance of general (ie non-cancer specific) PA programmes to them,⁴⁵ and there is
42 evidence that tailoring interventions to a specific target population is likely to increase
43 effectiveness.⁴⁶ Taken together, this suggests that the route most likely to lead to changes in CCSs'
44 PA levels is to develop an intervention specifically for this group.
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49 The final output from this phase of the BEACON study will be a prototype evidence-based and
50 theoretically-informed intervention. The next step will be to fully operationalise the intervention and
51 any supporting materials (e.g. training manual). Efficient and systematic user pre-testing studies will
52 be conducted to provide insight into different aspects of the intervention and iteratively refine and
53 optimise it.³⁷ Subsequently, as recommended in the area of PA research,⁴⁷ we plan to assess
54 feasibility and acceptability to users and, following that, evaluate effectiveness and cost-
55 effectiveness in a randomised controlled trial, with a parallel process evaluation.⁴⁸
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58 Various organisations, including the American Cancer Society⁴⁹ and Macmillan Cancer Support,⁵⁰
59 have produced PA recommendations for cancer survivors (of all ages). However, understanding
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3 remains limited on how best to support survivors to improve levels of PA and maintain changes.^{51 52}
4 The study described here – although it focuses on CCSs – provides an example of how to use a
5 behavioural science approach to develop a person-centred, evidence-based and theoretically-
6 informed PA intervention and, therefore, may be informative for those interested in systematically
7 developing PA interventions for other survivor groups.
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11

12 **Author contributions**

13
14 MB, LS, RS and VAS conceived the study. MB, LS, RS, VAS, NS and AG developed the grant proposal
15 and secured funding. MB managed the ethical and research approvals and will undertake the field
16 work and data analysis supported primarily by LS and VAS. RS, AG and NS will support recruitment of
17 participants. JH & OB have, and will continue to, provide PPI input. JS and KM will provide specialist
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34 **Figure 1:** Overview of planned research for the BEACON project

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36 **Table 1:** Domains, actions and planned methods to develop physical activity intervention for
37 childhood cancer survivors
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39 **Table 2:** Childhood cancer survivors’ inclusion and exclusion criteria
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Table 1: Domains, actions and planned methods to develop physical activity intervention for childhood cancer survivors

Key actions to consider for intervention development ¹⁶	Domain of intervention development and associated specific action(s) ¹⁵	Methods utilised, or planned in this research to develop intervention
Planning the process Involving stakeholders Bringing together a team	Conception	
	1. Identifying the problem in need of a new intervention (including the health problem, the problematic behaviour and the target population)	<ul style="list-style-type: none"> • Identification and evaluation of the literature on: <ul style="list-style-type: none"> - Prevalence of cardiovascular late effects in CCSs - Low PA in CCSs - Benefit of PA to the health of CCSs • Clinical experience/knowledge of low PA in CCSs.
	Planning	
	2. Setting up a planning group/development team	<ul style="list-style-type: none"> • Establishing a multidisciplinary steering group involving: <ul style="list-style-type: none"> - Researchers and academics with expertise in health psychology, behavioural science/intervention development, exercise physiology, PA interventions in clinical populations, digital health innovation. - HCPs and service providers (consultant oncologists, nurse specialists). - Patient representatives.
Reviewing published evidence Undertaking primary data collection Drawing on existing theories Articulate programme theory Understanding context Attending to future implementation	3. Understanding the problem to be addressed <i>i. Understanding the views and experiences and psycho-social of the potential target population</i>	<ul style="list-style-type: none"> • Use of patient and public involvement during initial stages of planning: <ul style="list-style-type: none"> - Gaining young peoples' views of PA and the study concept via an NHS young persons' advisory group - Consultation with CCSs on the need for PA interventions via focus group and survey methods • Identification of literature reporting HCPs (who may be involved in the resulting intervention) views of PA in CCSs, and the provision of PA advice to CCSs.
	<i>ii. Assessing the causes of the problems</i>	<ul style="list-style-type: none"> • Undertaking research to explore and understand CCSs views and experiences of PA <ul style="list-style-type: none"> - Literature review of the barriers and facilitators to PA in CCSs - Undertaking in-depth interviews with CCSs and their parents regarding their views and experiences of PA, including perceived barriers and facilitators (informed by the Theoretical Domains Framework) - Creating a logic model of the problem
	<i>iii. Describing and understanding the wider context of the target population and the context in which the intervention will be implemented</i>	<ul style="list-style-type: none"> • Undertaking research to explore the views of CCSs views of receiving lifestyle/PA advice in follow-up care. <ul style="list-style-type: none"> - Survey and interviews with CCSs attending follow-up care • Involving HCPs, service providers and patients in steering group and in co-production of intervention.
	<i>iv. Identifying the effectiveness of interventions for PA in CCSs</i>	<ul style="list-style-type: none"> • Identification and evaluation of existing PA interventions in CCSs. <ul style="list-style-type: none"> - Systematic reviews of PA interventions in CCSs - Research evaluating PA interventions in CCSs
	<i>v. Understanding wider stakeholders' perspectives of problems and issues</i>	<ul style="list-style-type: none"> • Actively engage with stakeholders, service providers and patients throughout research and in co-production of intervention.

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4	Drawing on existing theories Articulating programme theory Understanding context Attending to future implementation	4. <i>Making decisions about aims and goals of intervention</i>
5		Based on the evidence generated in WP1, the steering group and research team will make decisions on the specific aims and goals of intervention. These will be presented and discussed in co-design workshops.
6		5. <i>Identifying what needs to change, how to bring about change</i>
7		Following WP1 a logical model of change will be developed for PA in CCS drawing on the evidence and constructs from relevant theories.
8		6. <i>Specify who will change, how and when</i>
9		<ul style="list-style-type: none"> Following WP1 the steering group and research team will break down the behavioural outcomes to consider, prioritise and map who needs to change what, how changes will occur as a result of the intervention and when these changes are expected to take place.
10		<ul style="list-style-type: none"> Involvement of HCPs/service providers in development team Co-design workshops with stakeholders (including HCPs) Use of Normalisation Theory Process to inform discussion with HCPs
11		<ul style="list-style-type: none"> Steering group and stakeholder input on feasibility of intervention
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25	Designing and refining intervention	9. <i>Generating ideas about solutions and components and features of an intervention</i>
26		<ul style="list-style-type: none"> Mapping of behavioural determinants onto behaviour change techniques using Behaviour Change Wheel. Co-design workshops using creative methods and activities to enable idea generation
27		10. <i>Re-visit decisions about where to intervene</i>
28		<ul style="list-style-type: none"> Input from stakeholders to make final decisions regarding the scope, the target population, key features and components of intervention which will be further refined during workshops.
29		11. <i>Make decisions about the content, format and delivery of the intervention</i>
30		<ul style="list-style-type: none"> Findings of WP1 will be combined into a theoretical model of PA in CCSs and to develop initial ideas about content, format and delivery. Actively engaging with steering group and stakeholders via co-design workshops to obtain views on the potential content, format and delivery of intervention
31		<ul style="list-style-type: none"> Design of potential implementation plan will be informed by discussions with HCPs and other stakeholder regarding potential implementation barriers and previous research.
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46		13. <i>Make prototypes/mock-ups of the intervention where relevant</i>
47		<ul style="list-style-type: none"> Generation and discussion of mock-ups and paper-based prototypes during co-design workshop. Feedback
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Table 2: CCSs' inclusion and exclusion criteria

Inclusion criteria	Exclusion criteria
Diagnosed with any haematological malignancy or solid or CNS tumour under the age of 19 years	Any cognitive or physical impairment of sufficient severity to limit their ability to understand, engage with or undertake PA
Currently aged 10-24 years	Any contraindications to exercise
Currently 2-15 years from the end of treatment	
No active disease	

For peer review only

Input from steering group and PPI

Workpackage 1

In-depth interviews informed by theory

To identify perceived barriers and facilitators to physical activity.

Childhood cancer survivors recruited from two NHS trusts

Parents of childhood cancer survivors recruited from two NHS trusts

Thematic analysis of interview data

Previous primary research with childhood cancer survivors and systematic reviews of relevant literature will also inform work package 2

Workpackage 2

Phase 1: Behavioural analysis

Identifying potentially modifiable determinants of PA in CCSs and developing a theoretical model of PA in CCSs. Identifying potential intervention functions, components and behaviour change techniques.

Phase 2: Co-design process

Stage 1 co-design activities
Testing evidence statements.
Generate new ideas for interventions.

Participants:
CCSs, parents, HCPs, academics/researchers.

Output:
Intervention principles.

Stage 2 co-design activities
User feedback on intervention principles.
Develop core intervention ideas/concepts.
Design prototypes of potential interventions.

Participants:
CCSs, parents, HCPs, academics/researchers.

Output:
Core concepts.
Paper prototypes.

Final output: Prototype intervention

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Figure 1: Overview of planned research for the BEACON project

BMJ Open

Using qualitative and co-design methods to inform the development of an intervention to support and improve physical activity in childhood cancer survivors: a study protocol for BEing Active after ChildhOod caNcer (BEACON)

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Manuscript ID	bmjopen-2020-041073.R1
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Date Submitted by the Author:	02-Sep-2020
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Primary Subject Heading:	Oncology
Secondary Subject Heading:	Qualitative research
Keywords:	Protocols & guidelines < HEALTH SERVICES ADMINISTRATION & MANAGEMENT, Paediatric oncology < ONCOLOGY, QUALITATIVE RESEARCH

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3 **Using qualitative and co-design methods to inform the development of an intervention to support**
4 **and improve physical activity in childhood cancer survivors: a study protocol for BEing Active after**
5 **Childhood caNcer (BEACON)**
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ABSTRACT

Introduction:

Childhood cancer survivors (CCSs) treated with cardiotoxic cancer treatments are at increased risk of developing cardiometabolic complications. This risk is further exacerbated by poor health behaviours. In particular, CCSs are less active than non-cancer comparators. Existing interventions aiming to improve physical activity (PA) levels in CCSs are methodologically weak. The aim of this study is to rigorously and systematically develop an evidence- and theory-based intervention to promote, support, improve and sustain PA levels in CCSs, with the long-term goal of reducing CCSs' cardiovascular morbidity and mortality.

Methods and analysis:

The Being Active after Childhood Cancer (BEACON) study involves two workpackages at two NHS sites in England, UK. *Participants:* CCSs and their parents, and healthcare professionals (HCPs) involved in their care. *Methods:* Workpackage one (WP1) will use qualitative methods to explore and understand the barriers and facilitators to PA in CCSs. Two sets of semi-structured interviews will be conducted with (i) CCSs (aged 10 -24 years) and (ii) parents of CCSs. Workpackage two (WP2) will use co-design methods to bring together stakeholders (CCSs; their parents; HCPs; researchers) to develop a prototype intervention. Where possible, all data will be audio-recorded and transcribed. *Analysis:* Data from WP1 will be analysed using a thematic approach. Analysis of WP2 data will involve content analysis, and analysis of formative output and procedures.

Ethics and dissemination:

The study was approved by North East - Tyne & Wear South Research Ethics Committee (REC ref: 18/NE/0274). Research findings will be disseminated primarily via national and international conferences and publication in peer-reviewed journals. Patient and public involvement (PPI) will inform further dissemination activities.

ARTICLE SUMMARY

Strengths and limitations of this study

- The BEACON study will provide in-depth knowledge on the barriers and facilitators to PA in CCSs.
- The use of recognised frameworks of intervention development and principles from behavioural science to systematically develop an evidence- and theory-based health behaviour change intervention is a significant strength.
- Intervention development will actively engage stakeholders (CCSs, parents and healthcare professionals), ensuring that the resulting intervention is co-produced with those it aims to support, and maximizing likely acceptability and feasibility.
- Strategies will mitigate against potential sources of bias and challenges in recruitment. COVID-19 guidelines at the time of study, as well as participant preference, will dictate the

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3 modes of participation. As such we offer multiple ways in which individuals may contribute
4 to the study and recognise the potential limitations of conducting co-design work remotely.
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- 6 • The prototype intervention developed will be ready to be taken forward into production and
7 testing.
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10 11 INTRODUCTION

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13 As a result of treatment advances, the population of childhood cancer survivors (CCSs) has rapidly
14 grown. In the UK alone, there are more than 40,000 CCSs,¹ whilst across Europe there may be up to
15 500,000.² Two-thirds of CCSs may develop chronic health conditions by 15-25 years post-diagnosis.³
16 In particular, those treated with cardiotoxic therapies can experience persistent and cumulative
17 damage to their cardiovascular, pulmonary and metabolic systems.⁴ Cardiovascular complications
18 are a leading cause of morbidity among CCSs, and British CCSs have a 3.4-fold excess risk of cardiac
19 death.⁵ Development of these chronic conditions impacts adversely not only on the survivors'
20 physical health, but also on their psychological health and wellbeing and incurs costs for the
21 healthcare system.
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25 In the general population, it is well recognised that poor cardiovascular outcomes are strongly
26 related to modifiable health behaviours, including a lack of physical activity (PA).⁶ Similarly, low
27 levels of PA in even young CCSs (<18 years), have been linked to a worse cardiovascular risk profile,⁷
28 and CCSs are often less active than controls without a history of cancer.⁸ However, amongst long-
29 term survivors of childhood lymphoma, a higher levels of vigorous PA is associated with a 50% lower
30 risk of any cardiovascular event.⁹ Moreover, a Cochrane review indicated that physical exercise
31 training programmes may improve physical fitness, body composition and cardiorespiratory fitness
32 in childhood cancer patients and survivors.¹⁰ Considered together, this evidence provides a strong
33 rationale for developing effective interventions to increase PA in CCSs.
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37 Reviews have concluded that interventions to increase PA levels among CCSs are feasible and safe.
38 However, studies are heterogeneous and most are methodologically limited.^{11 12} In addition, there is
39 little evidence that interventions have been systematically developed using recognised frameworks
40 of intervention development.¹³ Critically, while an understanding of factors which may promote or
41 inhibit the target health behaviour (here, PA) is an essential first step in intervention design,¹⁴⁻¹⁷
42 most interventions appear to have been developed without having undertaken formative work to
43 gain this understanding. Indeed, currently, little is known about determinants of PA behaviours in
44 CCSs. Additionally, although the application of appropriate theory is recognised in behavioural
45 science as an essential element of behaviour change interventions,¹⁴⁻¹⁷ most interventions appear to
46 have no robust theoretical underpinnings.
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50 Leading authorities advocate active stakeholder involvement in the design and development of
51 novel health interventions,¹⁴⁻¹⁷; this is essential to understanding the perspectives and psychosocial
52 context of users.¹⁸ However, most interventions for CCSs have been developed without the
53 involvement of CCSs, raising concerns about the relevance and acceptability of the interventions to
54 survivors. Moreover, although parents are key agents in their children's PA behaviour,^{19 20} there has
55 been little attempt to understand either: (i) how the beliefs of parents of CCSs might influence their
56 child's PA behaviours, or (ii) how parental support may be harnessed to encourage PA in CCSs.
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59 A further consideration is wider implementation of interventions among CCSs. Those developed thus
60 far were not designed to be deliverable within the context of the UK National Health Service (NHS).

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3 More generally, there is a lack of research exploring how support to modify health behaviours
4 (including PA) among CCSs can be implemented effectively and feasibly in follow-up care.
5 Involvement of HCPs and other relevant stakeholders in the development process would increase
6 the likelihood that an intervention will be feasible, acceptable and implementable in the current
7 healthcare pathway for CCSs; this does not appear to have been widely done.
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10 This project seeks to comprehensively investigate barriers and enablers to PA among CCSs – from
11 the CCSs, parental and wider stakeholder perspective. Using the knowledge gained, and with the
12 support of key stakeholders, we will develop a person-centred evidence- and theory-based
13 prototype intervention aimed at promoting and supporting sustainable PA behaviour change in
14 CCSs.
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17 18 **AIMS AND OBJECTIVES**

19 20 **Aim**

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22 The ultimate goal of the BEACON project is to develop an intervention which can reduce
23 cardiometabolic risk markers in the medium-term, and reduce cardiovascular morbidity and
24 mortality in the long-term, whilst also helping to prevent a deterioration in patient well-being and
25 health-related quality of life due to poor cardiovascular health.
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28 29 **Objectives**

30 The objectives of this phase of the BEACON project are to:

- 31 1) explore CCSs' experiences of, and participation in, PA behaviours.
- 32 2) identify and explore the barriers and enablers of PA behaviours in CCSs.
- 33 3) explore CCSs' and parents' experiences of receiving advice on PA or exercise, and perceived
34 need for this information.
- 35 4) actively engage key stakeholders (CCSs, their parents, HCPs) in a co-design process to
36 develop a prototype intervention.
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43 44 **METHODS AND ANALYSIS**

45 46 **Study design**

47 The study will be informed by intervention development approaches - notably the Medical Research
48 Council framework of intervention development¹⁵ and the Person-Based Approach¹⁸ to ensure that
49 the resulting intervention is systematically developed from the bottom-up and: 1) is theory- and
50 evidence-based;^{14 15 17 21-23} 2) prioritises and incorporates the views of the people who will use the
51 intervention;^{18 24} and 3) is likely to be implementable and scalable in the NHS.^{25 26} The current phase
52 of the project is focused on the intervention development. The planned research activities (which
53 form two sequential workpackages), and the other formative work previously undertaken by the
54 study team which will feed into intervention development, are summarized in Table 1 and Figure 1.
55 Workpackage 1 (WP1) will generate evidence on determinants of PA among CCSs. Workpackage 2
56 (WP2) will involve a co-design process to produce a prototype intervention.
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Participants

WP1 involves CCSs and parents/guardians of CCSs; eligibility criteria are shown in Table 2. Participants will be recruited via two clinical sites which are both specialist centres in childhood cancer treatment.

WP2 will recruit CCSs, parents/guardians of CCSs and HCPs. The same inclusion and exclusion criteria as for WP1 will apply for CCSs and parents/guardians. HCPs will be eligible if they are involved in the follow-up care of CCSs. Academics and researchers with relevant expertise will also be eligible.

Sampling

WP1: Generating evidence on determinants of PA among CCSs

Potential participants will be selected using purposive sampling with strata comprising of: age (for CCSs - current age of 10-15/16-24 years; for parents/guardians - age of child at diagnosis: ≤ 10 ; 11-18 years); clinical site); and cancer site (haematological malignancy/solid tumour/central nervous system tumour). Diversity in other characteristics (e.g. gender, treatment, time since diagnosis) will be sought to ensure sample heterogeneity and elicitation of a broad range of views and experiences.

Recruitment will continue until data saturation is reached in each interview set, defined as no new themes arising in the last three interviews.²⁷ We estimate that, interviews with 25-30 CCSs and 25-30 parents/guardians will provide adequate data. CCSs may participate without their parent/guardian taking part, and vice versa.

WP2: Co-design process

Up to 40 CCSs, parents/guardians of survivors, HCPs and academics/researchers will take part in the co-design process. At least 20 participants will be CCSs (due to the nature of the activities, participation is deemed only to be suitable for CCSs aged 16 and above) and 10 will be parents/guardians. As with the interviews, it will be important to seek diversity in the participants.

HCPs invited to take part in WP2 will include consultant oncologists and nurse specialists from paediatric and teenage and young adult services and other relevant HCPs (e.g. occupational therapists, physiotherapists, psychologists, cardiologists). The aim is that at least six HCPs will participate in the co-design process. Academics/researchers with expertise in the following fields will also participate: behavioural science, health psychology, PA and exercise science, healthcare technologies and human-centred design, and childhood cancer survivorship.

Identification, screening and recruitment of sample

Identification and screening of CCSs began in February 2019 with recruitment expected to end December 2020.

WP1: Generating evidence on determinants of PA among CCSs

Employing multiple recruitment strategies can guard against recruitment problems,²⁸ therefore, we will use up to three methods for recruiting participants. The primary method will require consultant oncologists/nurse specialists to screen attendance lists of forthcoming CCSs follow-up clinics at

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3 collaborating sites. At the clinic, eligible CCSs and their parents/guardians will be informed of the
4 study by their child's oncologist/nurse specialist and asked if they would like to meet the researcher
5 (MB). If so, the researcher (MB) will provide further details including the study information sheet
6 and answer any questions. Potentially interested CCSs and parents/guardians will be asked whether
7 the researcher can contact them in a few days to find out whether they would like to participate.
8 Meeting the researcher at clinic will help potential participants feel more at ease and aid the
9 establishment of rapport, which may be particularly important for younger patients.²⁹
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12 Approvals are in place for variants of this process in the event that the researcher cannot attend the
13 clinic, or the clinic is too busy for collaborating clinical colleagues to approach eligible CCSs and
14 parents/guardians individually. These include (1) clinical colleagues recording details of those
15 potentially interested of behalf of the researcher, who will follow-up by phone and (2) provision of
16 study packs (containing a reply slip) to eligible CCSs and parents/guardians at clinic check-in with
17 those interested returning the reply slip in a sealed envelope to the receptionist to forward to the
18 researcher.
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21 The second method will involve consultant oncologists/nurse specialists at the two sites screening
22 patients in their care for eligibility and mailing a study information sheet; follow-up telephone calls
23 by the clinical colleagues are permitted. Interested CCSs and parents/guardian may contact the
24 researcher directly.
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27 The third method, if required, will be identification of survivors via cancer registries in the study
28 areas: the Northern Region Young Person's Malignant Disease Registry (NRYPMDR) and the
29 Yorkshire Specialist Register of Cancer in Children and Young People (YSRCCYP).
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31 Information sheets for CCSs are developmentally appropriate and designed for ages 10-12, 13-15
32 and ≥16 years. CCSs aged 10-15 years will also receive a copy of the Charter of Rights for Children
33 and Young People in Research.³⁰ Parents of CCSs aged 10-15 years will receive an information sheet
34 explaining the study their child has been invited to participate in.
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39 WP2: Co-design process

40 WP1 participants will be asked if they wish to be notified of/invited to the co-design activities. New
41 participants, without experience of the study, will also be recruited. Methods for recruiting CCSs and
42 their parents/guardians will mirror those of WP1. In addition, we will also seek to recruit via social
43 media, support groups and charities. Posts advertising the study will ask for interested individuals to
44 contact the researcher who will assess eligibility and provide further information.
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47 Eligible HCPs working at the collaborating sites will be invited to take part. We will also promote the
48 study via social media, and through networks of the Childhood Cancer & Leukaemia Group, to
49 encourage participation of HCPs from across UK. Academics and researchers experienced in relevant
50 areas (also previously stated) will also be asked to participate.
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56 **Data collection**

57 WP1: Generating evidence on determinants of PA among CCSs

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3 Interviews with CCSs aged 10-15 years will take place face-to-face at the interviewee's and their
4 parents preferred location (e.g. university/home). A parent/guardian may be present if they or the
5 child wishes. These interviews are expected to last 30-60 minutes, but length will be determined by
6 the child.
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8 Interviews with CCSs aged ≥ 16 years, and those with parents/guardians, will take place by
9 telephone, an end-to-end encrypted web app which enables secure audio/video calls (e.g.
10 WhatsApp, Zoom), or face-to-face at a location of their choosing; providing choice on ways to
11 participate can help maximise recruitment.²⁸ Experience suggests these interview will last 60-90
12 minutes³¹, but may be longer if the interviewee wishes.
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15 Before the interview commences, the researcher will seek informed consent; for those aged 10-15
16 years, a parent/guardian will provide consent and the interviewee assent to ensure that the child
17 feels involved in the decision about their participation.³² Participants (and parents on behalf of
18 children aged 10-15 years) will be asked to complete a short demographic questionnaire. Time will
19 be spent developing rapport and creating a secure, trusting environment,²⁸ particularly with younger
20 children.²⁹
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23 The interviews will be guided by a topic guide, which will be informed by the Theoretical Domains
24 Framework (TDF), an integrative framework of behaviour change theories,^{21 22} and will cover:
25 participant's views and attitudes towards their own/their child's PA; difficulties experienced with,
26 and barriers to, PA; whether support/advice has been given regarding PA; and what helps or would
27 help the survivor to be more active. Questions will be open and neutral. Topic guides will be used
28 flexibly to allow interviewees to raise issues they consider important to the topic (PA); if this results
29 in new areas, these will be explored in subsequent interviews to ensure sufficient depth is reached.
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32 Interview content will be developmentally appropriate. Interviews with children aged 10-15 years
33 will utilise cue cards and images to help engage and focus the participant and provide them with
34 some control over the order of the questions.²⁹ With the interviewees' permission, interviews will be
35 audio-recorded; if permission for recording is not granted, the researcher will take detailed notes.
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40 WP2: Co-design process

41 We will follow the sequential and systematic co-design approach to integrate scientific evidence,
42 expert knowledge and experience, and stakeholder involvement to design a prototype
43 intervention.²⁴ The two phases of WP2 are described below.
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48 *Phase 1: Behavioural analysis*

49 The researcher team will combine WP1 findings and other formative work previously conducted
50 (e.g. a systematic review) (Figure 1), into a "theoretical model" of PA engagement among CCSs. They
51 will identify which influences on PA are potentially modifiable to determine what needs to be done
52 to change behaviours. The Behaviour Change Wheel will be used to map the TDF domains, and
53 organise these into a working theoretical model of PA in CCSs.²³ For each identified factor the team
54 will identify which intervention functions might be effective in changing PA behaviours. For each of
55 the relevant intervention functions, associated behavioural change techniques will be identified (i.e.
56 the techniques that can be used to overcome barriers to, and enhance enablers of, engagement with
57 PA).³³
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Phase 2: Co-design process

This process will involve a range of methods in order to engage and collaborate with stakeholders flexibly. Due to the current global COVID-19 pandemic, we will offer multiple modes of participation including workshops (face-to-face or online), interviews (one-to-one or small groups of 2/3 people; face-to-face or online); and online collaborative groups. The mode of participation will be guided by participant preference, as well as COVID-19 guidelines at the time of study.

Face-to-face workshops are expected to last 3-4 hours, while those online will be shorter (1-2 hours). Interviews are likely to also last 1-2 hours. Video-conferencing is an acceptable method for discussion with young people and an optimal alternative to face-to-face groups; it also enables people from various geographical location to attend.³⁴

Online collaborative groups allow stakeholders to engage with the development process, and one another, both in real time and asynchronously. Secure groups will be set up via WhatsApp, Facebook or an Ideaboard.co.uk website developed specifically for the study. WhatsApp and Facebook are widely used and familiar applications, and have successfully been used for co-design^{35 36}; Ideaboard offers greater flexibility. Preferences of potential participants will inform the choice of platform. The team will post content to the groups (e.g. videos, images, questions) and invite feedback.

Following PPI input, CCSs will have the choice to participate in a survivor only or mixed (survivors plus parents/HCPs) workshop/small group interview/collaborative group.

Using these methods, a range of activities (e.g. think aloud, mapping, brainstorming, storyboarding) will be used to engage participants, provide ways for them to share, envision and develop their ideas with others and to facilitate interaction. The specific activities will be dictated by the findings of WP1, and the findings of any preceding workshops.

Two steps in the co-design process are envisioned (Figure 1). In the first step, evidence statements on PA among CCSs will be presented. Stakeholders' views on the relevance, importance and effectiveness will be sought. Activities will be used to generate insights into what is needed to improve PA levels in CCSs and novel intervention ideas which stakeholders think could be effective and acceptable in improving PA in CCSs. Ideas will also be sought for how an intervention should be designed, where and how it should be implemented, and the relevant components. Mapping activities will enable organisation and visualisation of resulting intervention ideas and their key components. The research team will analyse information collected to develop 'intervention principles', ensuring that the evidence and theory, which is central to the success of the intervention, remains intact.³⁷

During the second stage activities will focus on gaining user feedback on intervention principles. Content and mode of delivery will be further developed and refined. Intervention tailoring will also be considered. Participants will identify and discuss potential challenges around acceptability, usability and feasibility from different perspectives (e.g. CCSs, parents, HCPs, commissioners, service providers). Based on participants discussion and decisions, designers/creative facilitators will begin to sketch paper-based 'mock-ups' of the intervention.

Outputs will be critically evaluated and translated into a design brief which details the aims of the intervention, the design features it will include, and how these will be operationalised, taking care will to ensure alignment with evidence and theory. A logic model will be developed,¹⁴ providing a graphical/textual representation of how the intervention is intended to work, linking outcomes with

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3 processes, the underlying theoretical assumptions and active ingredients (or the behaviour change
4 methods and techniques that will be used to target the identified processes/mechanism associated
5 with behaviour and behaviour change).^{33,38} The outcome will be a mock-up of the prototype which
6 will represent the main features of the intervention. This prototype will be ready to take into
7 production and undergo refinement and optimisation before going forward into further testing in a
8 future study.
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11 Prior to participation in co-design activities, informed consent will be sought and a ground rules for
12 communication and engagement established. The researchers will ensure an atmosphere which is
13 welcoming and non-judgemental and will be clear that all participants are treated as equals whether
14 they are young people, parents, researchers or HCPs.
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16 Where possible, co-design activities will be audio or video recorded and transcribed. Other data
17 collected will include written data/notes, mapping activities and sketches resulting from the various
18 activities (e.g. group work, brainstorm) and written comments generated by the online collaborative
19 groups.
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22 23 24 **Data analysis plan**

25 WP1: Generating evidence on determinants of PA among CCSs

26 Interview recordings will be transcribed verbatim. Analysis will occur in parallel with data collection
27 to ensure that any new issues raised are explored in subsequent interviews. Interviews from CCSs
28 and parents/guardians will be analysed separately. To identify views and experiences of, and
29 barriers and facilitators to, PA in CCSs an inductive thematic analysis will be conducted.³⁹ Two team
30 members will code data from preliminary interviews and discuss and agree the emerging codes and
31 potential themes. Codes relating to the barriers and facilitators to PA will be mapped onto the TDF.²¹
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22 These codes will then be applied by the researcher to remaining interviews, incorporating any new
23 codes and themes as they are identified. For analytical rigour, the classification of belief statements
24 to the TDF domains will be discussed and agreed within the team. Coding and analysis will be
25 facilitated by QSR International's NVivo software (Version 12, 2018).

26 WP2: Co-design activities

27 Analysis of co-design activities will be focused specifically on the aims of each activity, pragmatic and
28 expeditious so findings can be fed into subsequent stages.³⁷ Qualitative content analysis will be
29 performed using QSR International's NVivo software (Version 12, 2018), supplemented by other
30 forms of analysis as required.
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51 **Ethics and dissemination**

52 A favourable opinion has been granted from the North East - Tyne & Wear South Research Ethics
53 Committee (REC ref: 18/NE/0274).
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56 Informed consent will be sought prior to participation. Participants will be informed that
57 participation is entirely voluntary, and they may withdraw at any point, without giving a reason and
58 without negative consequences. They will be asked for their agreement to audio/video record
59 (where relevant) and informed that recordings are confidential, and transcriptions of audio-
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3 recordings will be anonymised. Ethical considerations relating to the interviewing of children (aged
4 under 16 years) are described above. Interview participants will be offered payment of any travel
5 expenses and a £20 high-street shopping voucher. This amount is based on the need to provide
6 some compensation for the participant's time, expertise and contribution to the research but
7 without coercing individuals to take part when they would rather not.⁴⁰ Participants will be notified
8 of the voucher in the study information sheet and will be offered the voucher at the beginning of the
9 interview to convey to them that they are being rewarded for their attendance, and not for what
10 they share during the interview.⁴¹ Participants recruited to co-design activities will receive a high
11 street voucher which reflects the time commitment and nature of the activity they choose to
12 participate in, in accordance with INVOLVE guidance.⁴²

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14 Findings will be disseminated via our study website (<https://research.ncl.ac.uk/beingactive/>),
15 conferences and journal publication. A summary of research findings will be available for
16 participants. PPI will inform further dissemination activities (e.g. via patient organisations, social
17 media), appropriate formats (e.g. infographics, video) and content to ensure lay summaries are
18 understandable and engaging to survivors.

21 22 23 24 **Patient and public involvement and project steering group**

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26 Feedback on the study concept and methods were gained from two established PPI groups in
27 Newcastle: the Young Person's Advisory Group – North England (YPAG-NE) whose members are
28 young people aged 13-18 years old, and the Perspectives in Cancer Research group whose members
29 are survivors of adult cancer. Views of young adult CCSs were gained via representatives of two
30 European cancer organisations, PanCare (Pan-European Network for Care of Survivors after
31 Childhood and Adolescent Cancer) and Youth Cancer Europe, and a survey posted to a closed
32 Facebook group for survivors of cancer. YPAG-NE and two adult CCSs provided comments on patient
33 information and interview topic guides/cue cards. Patient representatives (OB & JH) sit on the
34 project steering group, and will be involved in data interpretation, co-design activities, and advising
35 on dissemination.

36 37 38 39 40 **DISCUSSION**

41
42 A high proportion of adolescents and young adults do not meet recommended levels of PA,⁴³ and
43 there are already many publicly available programmes and interventions to encourage PA, including
44 government/health service initiatives and apps.⁴⁴⁻⁴⁶ This raises the question of whether a specific PA
45 intervention is needed for CCSs. While there is considerable overlap between determinants of PA in
46 CCSs and young people without cancer, many influences are likely specific to CCSs (e.g. cancer-
47 related fatigue, frustration about impact of cancer).¹³ There are also concerns about the quality and
48 likely effectiveness of many of the publicly-available PA programmes.⁴⁶ In addition, cancer survivors
49 may question the relevance of general (i.e. non-cancer specific) PA programmes to them,⁴⁷ and
50 there is evidence that tailoring interventions to a specific target population is likely to increase
51 effectiveness.⁴⁸ Taken together, this suggests that the route most likely to lead to changes in CCSs'
52 PA levels is to develop an intervention specifically for this group.

53
54 The strength of our study lies in the adoption of an evidence-based, person-centred approach.
55 However, we also recognise the need to mitigate potential study limitations. To minimise selection
56 bias, the importance of giving all eligible patients the opportunity to hear about the study, and
57 allowing them make their own choice as to whether they want to participate or not, will be

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3 highlighted to those involved in the screening process.⁴⁹ For example, participating in this research
4 may appeal more to CCSs who are physically active, than to those who are not. Therefore, patients
5 will also be made aware that a judgement will not be made on their current activity levels and that
6 we are interested in their views regardless of whether they consider themselves to be active or not.
7 CCSs can be challenging to recruit to research,⁵⁰ therefore, we propose several routes by which CCSs
8 may be made aware of the study. This will help to safeguard against any potential recruitment issues
9 and will also ensure that a wide range of individuals are offered the opportunity to participate. We
10 also acknowledge the potential impact of the current COVID-19 pandemic on the study, including
11 the possible need to conduct co-design activities remotely. The use of video-conferencing could
12 exclude those who have limited access to the required technology, or those who do not feel
13 comfortable using it.⁵¹ The use of remote methods may also hinder the interactive, creative and
14 collaborative process essential to co-design. Therefore, we have proposed several ways that
15 individuals can take part in co-design activities, including online synchronous and asynchronous
16 methods. Careful and considered planning will be needed to adapt co-design activities to ensure
17 participation and engagement, as well as an online environment in which individuals feel safe and
18 able to contribute.

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22 The final output from this phase of the BEACON study will be a prototype evidence-based and
23 theoretically-informed intervention. The next step will be to fully operationalise the intervention and
24 any supporting materials (e.g. training manual). Efficient and systematic user pre-testing studies will
25 be conducted to provide insight into different aspects of the intervention and iteratively refine and
26 optimise it.³⁷ Subsequently, as recommended in the area of PA research,⁵² we plan to assess
27 feasibility and acceptability to users and, following that, evaluate effectiveness and cost-
28 effectiveness in a randomised controlled trial, with a parallel process evaluation.⁵³

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31 Various organisations, including the American Cancer Society⁵⁴ and Macmillan Cancer Support,⁵⁵
32 have produced PA recommendations for cancer survivors (of all ages). However, understanding
33 remains limited on how best to support survivors to improve levels of PA and maintain changes.^{56 57}
34 The study described here – although it focuses on CCSs – provides an example of how to use a
35 behavioural science approach to develop a person-centred, evidence-based and theoretically-
36 informed PA intervention and, therefore, may be informative for those interested in systematically
37 developing PA interventions for other survivor groups.

38 39 40 41 **Author contributions**

42 MB, LS, RS and VAS conceived the study. MB, LS, RS, VAS, NS and AG developed the grant proposal
43 and secured funding. MB managed the ethical and research approvals and will undertake the field
44 work and data analysis supported primarily by LS and VAS. RS, AG and NS will support recruitment of
45 participants. JH & OB have, and will continue to, provide PPI input. JS and KM will provide specialist
46 advice and expertise throughout the study. MB drafted the manuscript, and AG, JS, LS, RS, KM, NS,
47 VAS provided revisions and comments. All authors approved the final manuscript.

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51 number CCLGA 2017 26, the North of England Children's Cancer Research Fund and the Children's
52 Cancer Fund.

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57 **Competing interests statement:** No interests to declare.
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3 **Figure 1:** Overview of planned research for the BEACON project
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5 **Table 1:** Domains, actions and planned methods to develop physical activity intervention for
6 childhood cancer survivors
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8 **Table 2:** Childhood cancer survivors' inclusion and exclusion criteria
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For peer review only

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Table 1: Domains, actions and planned methods to develop physical activity intervention for childhood cancer survivors

Key actions to consider for intervention development ¹⁶	Domain of intervention development and associated specific action(s) ¹⁵	Methods utilised, or planned in this research to develop intervention
Planning the process Involving stakeholders Bringing together a team	Conception	
	1. Identifying the problem in need of a new intervention (including the health problem, the problematic behaviour and the target population)	<ul style="list-style-type: none"> • Identification and evaluation of the literature on: <ul style="list-style-type: none"> - Prevalence of cardiovascular late effects in CCSs - Low PA in CCSs - Benefit of PA to the health of CCSs • Clinical experience/knowledge of low PA in CCSs.
	Planning	
	2. Setting up a planning group/development team	<ul style="list-style-type: none"> • Establishing a multidisciplinary steering group involving: <ul style="list-style-type: none"> - Researchers and academics with expertise in health psychology, behavioural science/intervention development, exercise physiology, PA interventions in clinical populations, digital health innovation. - HCPs and service providers (consultant oncologists, nurse specialists). - Patient representatives.
Reviewing published evidence Undertaking primary data collection Drawing on existing theories Articulate programme theory Understanding context Attending to future implementation	3. Understanding the problem to be addressed <i>i. Understanding the views and experiences and psycho-social of the potential target population</i>	<ul style="list-style-type: none"> • Use of patient and public involvement during initial stages of planning: <ul style="list-style-type: none"> - Gaining young peoples' views of PA and the study concept via an NHS young persons' advisory group - Consultation with CCSs on the need for PA interventions via focus group and survey methods • Identification of literature reporting HCPs (who may be involved in the resulting intervention) views of PA in CCSs, and the provision of PA advice to CCSs.
	<i>ii. Assessing the causes of the problems</i>	<ul style="list-style-type: none"> • Undertaking research to explore and understand CCSs views and experiences of PA <ul style="list-style-type: none"> - Literature review of the barriers and facilitators to PA in CCSs - Undertaking in-depth interviews with CCSs and their parents regarding their views and experiences of PA, including perceived barriers and facilitators (informed by the Theoretical Domains Framework) - Creating a logic model of the problem
	<i>iii. Describing and understanding the wider context of the target population and the context in which the intervention will be implemented</i>	<ul style="list-style-type: none"> • Undertaking research to explore the views of CCSs views of receiving lifestyle/PA advice in follow-up care. <ul style="list-style-type: none"> - Survey and interviews with CCSs attending follow-up care • Involving HCPs, service providers and patients in steering group and in co-production of intervention.
	<i>iv. Identifying the effectiveness of interventions for PA in CCSs</i>	<ul style="list-style-type: none"> • Identification and evaluation of existing PA interventions in CCSs. <ul style="list-style-type: none"> - Systematic reviews of PA interventions in CCSs - Research evaluating PA interventions in CCSs

	v. <i>Understanding wider stakeholders' perspectives of problems and issues</i>	<ul style="list-style-type: none"> Actively engage with stakeholders, service providers and patients throughout research and in co-production of intervention.
Drawing on existing theories Articulating programme theory Understanding context Attending to future implementation	4. <i>Making decisions about aims and goals of intervention</i>	Based on the evidence generated in WP1, the steering group and research team will make decisions on the specific aims and goals of intervention. These will be presented and discussed in co-design workshops.
	5. <i>Identifying what needs to change, how to bring about change</i>	Following WP1 a logical model of change will be developed for PA in CCS drawing on the evidence and constructs from relevant theories.
	6. <i>Specify who will change, how and when</i>	<ul style="list-style-type: none"> Following WP1 the steering group and research team will break down the behavioural outcomes to consider, prioritise and map who needs to change what, how changes will occur as a result of the intervention and when these changes are expected to take place.
	7. <i>Considering the real-world issues about cost and delivery of any intervention to reduce risk of implementation failure</i>	<ul style="list-style-type: none"> Involvement of HCPs/service providers in development team Co-design workshops with stakeholders (including HCPs) Use of Normalisation Theory Process to inform discussion with HCPs
	8. <i>Considering whether it is worthwhile continuing with development of intervention</i>	<ul style="list-style-type: none"> Steering group and stakeholder input on feasibility of intervention
Designing and refining intervention	Designing	
	9. <i>Generating ideas about solutions and components and features of an intervention</i>	<ul style="list-style-type: none"> Mapping of behavioural determinants onto behaviour change techniques using Behaviour Change Wheel. Co-design workshops using creative methods and activities to enable idea generation
	10. <i>Re-visit decisions about where to intervene</i>	<ul style="list-style-type: none"> Input from stakeholders to make final decisions regarding the scope, the target population, key features and components of intervention which will be further refined during workshops.
	11. <i>Make decisions about the content, format and delivery of the intervention</i>	<ul style="list-style-type: none"> Findings of WP1 will be combined into a theoretical model of PA in CCSs and to develop initial ideas about content, format and delivery. Actively engaging with steering group and stakeholders via co-design workshops to obtain views on the potential content, format and delivery of intervention
	12. <i>Design an implementation plan, thinking about who will adopt the intervention and maintain it</i>	<ul style="list-style-type: none"> Design of potential implementation plan will be informed by discussions with HCPs and other stakeholder regarding potential implementation barriers and previous research.
	Creating	
	13. <i>Make prototypes/mock-ups of the intervention where relevant</i>	<ul style="list-style-type: none"> Generation and discussion of mock-ups and paper-based prototypes during co-design workshop. Feedback

Table 2: Childhood cancer survivors' inclusion and exclusion criteria

Inclusion criteria	Exclusion criteria
Diagnosed with any haematological malignancy or solid or CNS tumour under the age of 19 years	Any cognitive or physical impairment of sufficient severity to limit their ability to understand, engage with or undertake PA
Currently aged 10-24 years	Any contraindications to exercise
Currently 2-15 years from the end of treatment	
No active disease	

For peer review only

Input from steering group and PPI

Workpackage 1

In-depth interviews informed by theory

To identify perceived barriers and facilitators to physical activity.

Childhood cancer survivors (CCSs) recruited from two NHS trusts

Parents of CCSs recruited from two NHS trusts

Thematic analysis of interview data

Previous primary research with childhood cancer survivors and systematic reviews of relevant literature will also inform work package 2

Workpackage 2

Phase 1: Behavioural analysis

Identifying potentially modifiable determinants of PA in CCSs and developing a theoretical model of PA in CCSs. Identifying potential intervention functions, components and behaviour change techniques.

Phase 2: Co-design process

Stage 1 co-design activities
Testing evidence statements.
Generate new ideas for interventions.

Participants:
CCSs, parents, HCPs, academics/researchers.

Output:
Intervention principles.

Stage 2 co-design activities
User feedback on intervention principles.
Develop core intervention ideas/concepts.
Design prototypes of potential interventions.

Participants:
CCSs, parents, HCPs, academics/researchers.

Output:
Core concepts.
Paper prototypes.

Final output: Prototype intervention

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Figure 1: Overview of planned research for the BEACON project

BMJ Open

Using qualitative and co-design methods to inform the development of an intervention to support and improve physical activity in childhood cancer survivors: a study protocol for BEing Active after ChildhOod caNcer (BEACON)

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Manuscript ID	bmjopen-2020-041073.R2
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Date Submitted by the Author:	02-Nov-2020
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Primary Subject Heading:	Oncology
Secondary Subject Heading:	Qualitative research
Keywords:	Protocols & guidelines < HEALTH SERVICES ADMINISTRATION & MANAGEMENT, Paediatric oncology < ONCOLOGY, QUALITATIVE RESEARCH

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3 **Using qualitative and co-design methods to inform the development of an intervention to support**
4 **and improve physical activity in childhood cancer survivors: a study protocol for BEing Active after**
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60

ABSTRACT

Introduction:

Childhood cancer survivors (CCSs) treated with cardiotoxic cancer treatments are at increased risk of developing cardiometabolic complications. This risk is further exacerbated by poor health behaviours. In particular, CCSs are less active than non-cancer comparators. Existing interventions aiming to improve physical activity (PA) levels in CCSs are methodologically weak. The aim of this study is to rigorously and systematically develop an evidence- and theory-based intervention to promote, support, improve and sustain PA levels in CCSs, with the long-term goal of reducing CCSs' cardiovascular morbidity and mortality.

Methods and analysis:

The Being Active after Childhood Cancer (BEACON) study involves two workpackages at two NHS sites in England, UK. *Participants:* CCSs and their parents, and healthcare professionals (HCPs) involved in their care. *Methods:* Workpackage one (WP1) will use qualitative methods to explore and understand the barriers and facilitators to PA in CCSs. Two sets of semi-structured interviews will be conducted with (i) CCSs (aged 10 -24 years) and (ii) parents of CCSs. Workpackage two (WP2) will use co-design methods to bring together stakeholders (CCSs; their parents; HCPs; researchers) to develop a prototype intervention. Where possible, all data will be audio-recorded and transcribed. *Analysis:* Data from WP1 will be analysed using a thematic approach. Analysis of WP2 data will involve content analysis, and analysis of formative output and procedures.

Ethics and dissemination:

The study was approved by North East - Tyne & Wear South Research Ethics Committee (REC ref: 18/NE/0274). Research findings will be disseminated primarily via national and international conferences and publication in peer-reviewed journals. Patient and public involvement (PPI) will inform further dissemination activities.

ARTICLE SUMMARY

Strengths and limitations of this study

- The BEACON study will provide in-depth knowledge on the barriers and facilitators to PA in CCSs.
- The use of recognised frameworks of intervention development and principles from behavioural science to systematically develop an evidence- and theory-based health behaviour change intervention is a significant strength.
- Intervention development will actively engage stakeholders (CCSs, parents and healthcare professionals), ensuring that the resulting intervention is co-produced with those it aims to support, and maximizing likely acceptability and feasibility.
- Strategies will mitigate against potential sources of bias and challenges in recruitment. COVID-19 guidelines at the time of study, as well as participant preference, will dictate the

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3 modes of participation. As such we offer multiple ways in which individuals may contribute
4 to the study and recognise the potential limitations of conducting co-design work remotely.
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- 6 • The prototype intervention developed will be ready to be taken forward into production and
7 testing.
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10 11 INTRODUCTION

12
13 As a result of treatment advances, the population of childhood cancer survivors (CCSs) has rapidly
14 grown. In the UK alone, there are more than 40,000 CCSs,¹ whilst across Europe there may be up to
15 500,000.² Two-thirds of CCSs may develop chronic health conditions by 15-25 years post-diagnosis.³
16 In particular, those treated with cardiotoxic therapies can experience persistent and cumulative
17 damage to their cardiovascular, pulmonary and metabolic systems.⁴ Cardiovascular complications
18 are a leading cause of morbidity among CCSs, and British CCSs have a 3.4-fold excess risk of cardiac
19 death.⁵ Development of these chronic conditions impacts adversely not only on the survivors'
20 physical health, but also on their psychological health and wellbeing and incurs costs for the
21 healthcare system.
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25 In the general population, it is well recognised that poor cardiovascular outcomes are strongly
26 related to modifiable health behaviours, including a lack of physical activity (PA).⁶ Similarly, low
27 levels of PA in even young CCSs (<18 years), have been linked to a worse cardiovascular risk profile,⁷
28 and CCSs are often less active than controls without a history of cancer.⁸ However, amongst long-
29 term survivors of childhood lymphoma, a higher levels of vigorous PA is associated with a 50% lower
30 risk of any cardiovascular event.⁹ Moreover, a Cochrane review indicated that physical exercise
31 training programmes may improve physical fitness, body composition and cardiorespiratory fitness
32 in childhood cancer patients and survivors.¹⁰ Considered together, this evidence provides a strong
33 rationale for developing effective interventions to increase PA in CCSs.
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37 Reviews have concluded that interventions to increase PA levels among CCSs are feasible and safe.
38 However, studies are heterogeneous and most are methodologically limited.^{11 12} In addition, there is
39 little evidence that interventions have been systematically developed using recognised frameworks
40 of intervention development.¹³ Critically, while an understanding of factors which may promote or
41 inhibit the target health behaviour (here, PA) is an essential first step in intervention design,¹⁴⁻¹⁷
42 most interventions appear to have been developed without having undertaken formative work to
43 gain this understanding. Indeed, currently, little is known about determinants of PA behaviours in
44 CCSs. Additionally, although the application of appropriate theory is recognised in behavioural
45 science as an essential element of behaviour change interventions,¹⁴⁻¹⁷ most interventions appear to
46 have no robust theoretical underpinnings.
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50 Leading authorities advocate active stakeholder involvement in the design and development of
51 novel health interventions,¹⁴⁻¹⁷; this is essential to understanding the perspectives and psychosocial
52 context of users.¹⁸ However, most interventions for CCSs have been developed without the
53 involvement of CCSs, raising concerns about the relevance and acceptability of the interventions to
54 survivors. Moreover, although parents are key agents in their children's PA behaviour,^{19 20} there has
55 been little attempt to understand either: (i) how the beliefs of parents of CCSs might influence their
56 child's PA behaviours, or (ii) how parental support may be harnessed to encourage PA in CCSs.
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59 A further consideration is wider implementation of interventions among CCSs. Those developed thus
60 far were not designed to be deliverable within the context of the UK National Health Service (NHS).

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3 More generally, there is a lack of research exploring how support to modify health behaviours
4 (including PA) among CCSs can be implemented effectively and feasibly in follow-up care.
5 Involvement of HCPs and other relevant stakeholders in the development process would increase
6 the likelihood that an intervention will be feasible, acceptable and implementable in the current
7 healthcare pathway for CCSs; this does not appear to have been widely done.
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10 This project seeks to comprehensively investigate barriers and enablers to PA among CCSs – from
11 the CCSs, parental and wider stakeholder perspective. Using the knowledge gained, and with the
12 support of key stakeholders, we will develop a person-centred evidence- and theory-based
13 prototype intervention aimed at promoting and supporting sustainable PA behaviour change in
14 CCSs.
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17 18 **AIMS AND OBJECTIVES**

19 20 **Aim**

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22 The ultimate goal of the BEACON project is to develop an intervention which can reduce
23 cardiometabolic risk markers in the medium-term, and reduce cardiovascular morbidity and
24 mortality in the long-term, whilst also helping to prevent a deterioration in patient well-being and
25 health-related quality of life due to poor cardiovascular health.
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28 29 **Objectives**

30 The objectives of this phase of the BEACON project are to:

- 31 1) explore CCSs' experiences of, and participation in, PA behaviours.
- 32 2) identify and explore the barriers and enablers of PA behaviours in CCSs.
- 33 3) explore CCSs' and parents' experiences of receiving advice on PA or exercise, and perceived
34 need for this information.
- 35 4) actively engage key stakeholders (CCSs, their parents, HCPs) in a co-design process to
36 develop a prototype intervention.
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43 44 **METHODS AND ANALYSIS**

45 46 **Study design**

47 The study will be informed by intervention development approaches - notably the Medical Research
48 Council framework of intervention development¹⁵ and the Person-Based Approach¹⁸ to ensure that
49 the resulting intervention is systematically developed from the bottom-up and: 1) is theory- and
50 evidence-based;^{14 15 17 21-23} 2) prioritises and incorporates the views of the people who will use the
51 intervention;^{18 24} and 3) is likely to be implementable and scalable in the NHS.^{25 26} The current phase
52 of the project is focused on the intervention development. The planned research activities (which
53 form two sequential workpackages), and the other formative work previously undertaken by the
54 study team which will feed into intervention development, are summarized in Table 1 and Figure 1.
55 Workpackage 1 (WP1) will generate evidence on determinants of PA among CCSs. Workpackage 2
56 (WP2) will involve a co-design process to produce a prototype intervention.
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Participants

WP1 involves CCSs and parents/guardians of CCSs; eligibility criteria are shown in Table 2. Participants will be recruited via two clinical sites which are both specialist centres in childhood cancer treatment.

WP2 will recruit CCSs, parents/guardians of CCSs and HCPs. The same inclusion and exclusion criteria as for WP1 will apply for CCSs and parents/guardians. HCPs will be eligible if they are involved in the follow-up care of CCSs. Academics and researchers with relevant expertise will also be eligible.

Sampling

WP1: Generating evidence on determinants of PA among CCSs

Potential participants will be selected using purposive sampling with strata comprising of: age (for CCSs - current age of 10-15/16-24 years; for parents/guardians - age of child at diagnosis: ≤ 10 ; 11-18 years); clinical site); and cancer site (haematological malignancy/solid tumour/central nervous system tumour). Diversity in other characteristics (e.g. gender, treatment, time since diagnosis) will be sought to ensure sample heterogeneity and elicitation of a broad range of views and experiences.

Recruitment will continue until data saturation is reached in each interview set, defined as no new themes arising in the last three interviews.²⁷ We estimate that, interviews with 25-30 CCSs and 25-30 parents/guardians will provide adequate data. CCSs may participate without their parent/guardian taking part, and vice versa.

WP2: Co-design process

Up to 40 CCSs, parents/guardians of survivors, HCPs and academics/researchers will take part in the co-design process. At least 20 participants will be CCSs (due to the nature of the activities, participation is deemed only to be suitable for CCSs aged 16 and above) and 10 will be parents/guardians. As with the interviews, it will be important to seek diversity in the participants.

HCPs invited to take part in WP2 will include consultant oncologists and nurse specialists from paediatric and teenage and young adult services and other relevant HCPs (e.g. occupational therapists, physiotherapists, psychologists, cardiologists). The aim is that at least six HCPs will participate in the co-design process. Academics/researchers with expertise in the following fields will also participate: behavioural science, health psychology, PA and exercise science, healthcare technologies and human-centred design, and childhood cancer survivorship.

Identification, screening and recruitment of sample

Identification and screening of CCSs began in February 2019 with recruitment expected to end December 2020.

WP1: Generating evidence on determinants of PA among CCSs

Employing multiple recruitment strategies can guard against recruitment problems,²⁸ therefore, we will use up to three methods for recruiting participants. The primary method will require consultant oncologists/nurse specialists to screen attendance lists of forthcoming CCSs follow-up clinics at

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3 collaborating sites. At the clinic, eligible CCSs and their parents/guardians will be informed of the
4 study by their child's oncologist/nurse specialist and asked if they would like to meet the researcher
5 (MB). If so, the researcher (MB) will provide further details including the study information sheet
6 and answer any questions. Potentially interested CCSs and parents/guardians will be asked whether
7 the researcher can contact them in a few days to find out whether they would like to participate.
8 Meeting the researcher at clinic will help potential participants feel more at ease and aid the
9 establishment of rapport, which may be particularly important for younger patients.²⁹
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12 Approvals are in place for variants of this process in the event that the researcher cannot attend the
13 clinic, or the clinic is too busy for collaborating clinical colleagues to approach eligible CCSs and
14 parents/guardians individually. These include (1) clinical colleagues recording details of those
15 potentially interested of behalf of the researcher, who will follow-up by phone and (2) provision of
16 study packs (containing a reply slip) to eligible CCSs and parents/guardians at clinic check-in with
17 those interested returning the reply slip in a sealed envelope to the receptionist to forward to the
18 researcher.
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21 The second method will involve consultant oncologists/nurse specialists at the two sites screening
22 patients in their care for eligibility and mailing a study information sheet; follow-up telephone calls
23 by the clinical colleagues are permitted. Interested CCSs and parents/guardian may contact the
24 researcher directly.
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27 The third method, if required, will be identification of survivors via cancer registries in the study
28 areas: the Northern Region Young Person's Malignant Disease Registry (NRYPMDR) and the
29 Yorkshire Specialist Register of Cancer in Children and Young People (YSRCCYP).
30

31 Information sheets for CCSs are developmentally appropriate and designed for ages 10-12, 13-15
32 and ≥16 years. CCSs aged 10-15 years will also receive a copy of the Charter of Rights for Children
33 and Young People in Research.³⁰ Parents of CCSs aged 10-15 years will receive an information sheet
34 explaining the study their child has been invited to participate in.
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39 WP2: Co-design process

40 WP1 participants will be asked if they wish to be notified of/invited to the co-design activities. New
41 participants, without experience of the study, will also be recruited. Methods for recruiting CCSs and
42 their parents/guardians will mirror those of WP1. In addition, we will also seek to recruit via social
43 media, support groups and charities. Posts advertising the study will ask for interested individuals to
44 contact the researcher who will assess eligibility and provide further information.
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47 Eligible HCPs working at the collaborating sites will be invited to take part. We will also promote the
48 study via social media, and through networks of the Childhood Cancer & Leukaemia Group, to
49 encourage participation of HCPs from across UK. Academics and researchers experienced in relevant
50 areas (also previously stated) will also be asked to participate.
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56 **Data collection**

58 WP1: Generating evidence on determinants of PA among CCSs

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3 Interviews with CCSs aged 10-15 years will take place face-to-face at the interviewee's and their
4 parents preferred location (e.g. university/home). A parent/guardian may be present if they or the
5 child wishes. These interviews are expected to last 30-60 minutes, but length will be determined by
6 the child.
7

8 Interviews with CCSs aged ≥ 16 years, and those with parents/guardians, will take place by
9 telephone, an end-to-end encrypted web app which enables secure audio/video calls (e.g.
10 WhatsApp, Zoom), or face-to-face at a location of their choosing; providing choice on ways to
11 participate can help maximise recruitment.²⁸ Experience suggests these interview will last 60-90
12 minutes³¹, but may be longer if the interviewee wishes.
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15 Before the interview commences, the researcher will seek informed consent; for those aged 10-15
16 years, a parent/guardian will provide consent and the interviewee assent to ensure that the child
17 feels involved in the decision about their participation.³² Participants (and parents on behalf of
18 children aged 10-15 years) will be asked to complete a short demographic questionnaire. Time will
19 be spent developing rapport and creating a secure, trusting environment,²⁸ particularly with younger
20 children.²⁹
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23 The interviews will be guided by a topic guide, which will be informed by the Theoretical Domains
24 Framework (TDF), an integrative framework of behaviour change theories,^{21 22} and will cover:
25 participant's views and attitudes towards their own/their child's PA; difficulties experienced with,
26 and barriers to, PA; whether support/advice has been given regarding PA; and what helps or would
27 help the survivor to be more active. Questions will be open and neutral. Topic guides will be used
28 flexibly to allow interviewees to raise issues they consider important to the topic (PA); if this results
29 in new areas, these will be explored in subsequent interviews to ensure sufficient depth is reached.
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32 Interview content will be developmentally appropriate. Interviews with children aged 10-15 years
33 will utilise cue cards and images to help engage and focus the participant and provide them with
34 some control over the order of the questions.²⁹ With the interviewees' permission, interviews will be
35 audio-recorded; if permission for recording is not granted, the researcher will take detailed notes.
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40 WP2: Co-design process

41 We will follow the sequential and systematic co-design approach to integrate scientific evidence,
42 expert knowledge and experience, and stakeholder involvement to design a prototype
43 intervention.²⁴ The two phases of WP2 are described below.
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48 *Phase 1: Behavioural analysis*

49 The researcher team will combine WP1 findings and other formative work previously conducted
50 (e.g. a systematic review) (Figure 1), into a "theoretical model" of PA engagement among CCSs. They
51 will identify which influences on PA are potentially modifiable to determine what needs to be done
52 to change behaviours. The Behaviour Change Wheel will be used to map the TDF domains, and
53 organise these into a working theoretical model of PA in CCSs.²³ For each identified factor the team
54 will identify which intervention functions might be effective in changing PA behaviours. For each of
55 the relevant intervention functions, associated behavioural change techniques will be identified (i.e.
56 the techniques that can be used to overcome barriers to, and enhance enablers of, engagement with
57 PA).³³
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Phase 2: Co-design process

This process will involve a range of methods in order to engage and collaborate with stakeholders flexibly. Due to the current global COVID-19 pandemic, we will offer multiple modes of participation including workshops (face-to-face or online), interviews (one-to-one or small groups of 2/3 people; face-to-face or online); and online collaborative groups. The mode of participation will be guided by participant preference, as well as COVID-19 guidelines at the time of study.

Face-to-face workshops are expected to last 3-4 hours, while those online will be shorter (1-2 hours). Interviews are likely to also last 1-2 hours. Video-conferencing is an acceptable method for discussion with young people and an optimal alternative to face-to-face groups; it also enables people from various geographical location to attend.³⁴

Online collaborative groups allow stakeholders to engage with the development process, and one another, both in real time and asynchronously. Secure groups will be set up via WhatsApp, Facebook or an Ideaboard.co.uk website developed specifically for the study. WhatsApp and Facebook are widely used and familiar applications, and have successfully been used for co-design^{35 36}; Ideaboard offers greater flexibility. Preferences of potential participants will inform the choice of platform. The team will post content to the groups (e.g. videos, images, questions) and invite feedback.

Following PPI input, CCSs will have the choice to participate in a survivor only or mixed (survivors plus parents/HCPs) workshop/small group interview/collaborative group.

Using these methods, a range of activities (e.g. think aloud, mapping, brainstorming, storyboarding) will be used to engage participants, provide ways for them to share, envision and develop their ideas with others and to facilitate interaction. The specific activities will be dictated by the findings of WP1, and the findings of any preceding workshops.

Two steps in the co-design process are envisioned (Figure 1). In the first step, evidence statements on PA among CCSs will be presented. Stakeholders' views on the relevance, importance and effectiveness will be sought. Activities will be used to generate insights into what is needed to improve PA levels in CCSs and novel intervention ideas which stakeholders think could be effective and acceptable in improving PA in CCSs. Ideas will also be sought for how an intervention should be designed, where and how it should be implemented, and the relevant components. Mapping activities will enable organisation and visualisation of resulting intervention ideas and their key components. The research team will analyse information collected to develop 'intervention principles', ensuring that the evidence and theory, which is central to the success of the intervention, remains intact.³⁷

During the second stage activities will focus on gaining user feedback on intervention principles. Content and mode of delivery will be further developed and refined. Intervention tailoring will also be considered. Participants will identify and discuss potential challenges around acceptability, usability and feasibility from different perspectives (e.g. CCSs, parents, HCPs, commissioners, service providers). Based on participants discussion and decisions, designers/creative facilitators will begin to sketch paper-based 'mock-ups' of the intervention.

Outputs will be critically evaluated and translated into a design brief which details the aims of the intervention, the design features it will include, and how these will be operationalised, taking care will to ensure alignment with evidence and theory. A logic model will be developed,¹⁴ providing a graphical/textual representation of how the intervention is intended to work, linking outcomes with

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3 processes, the underlying theoretical assumptions and active ingredients (or the behaviour change
4 methods and techniques that will be used to target the identified processes/mechanism associated
5 with behaviour and behaviour change).^{33 38} The outcome will be a mock-up of the prototype which
6 will represent the main features of the intervention. This prototype will be ready to take into
7 production and undergo refinement and optimisation before going forward into further testing in a
8 future study.
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11 Prior to participation in co-design activities, informed consent will be sought and a ground rules for
12 communication and engagement established. The researchers will ensure an atmosphere which is
13 welcoming and non-judgemental and will be clear that all participants are treated as equals whether
14 they are young people, parents, researchers or HCPs.
15

16 Where possible, co-design activities will be audio or video recorded and transcribed. Other data
17 collected will include written data/notes, mapping activities and sketches resulting from the various
18 activities (e.g. group work, brainstorm) and written comments generated by the online collaborative
19 groups.
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22 23 24 **Data analysis plan**

25 WP1: Generating evidence on determinants of PA among CCSs

26 Interview recordings will be transcribed verbatim. Analysis will occur in parallel with data collection
27 to ensure that any new issues raised are explored in subsequent interviews. Interviews from CCSs
28 and parents/guardians will be analysed separately. To identify views and experiences of, and
29 barriers and facilitators to, PA in CCSs an inductive thematic analysis will be conducted.³⁹ Two team
30 members will code data from preliminary interviews and discuss and agree the emerging codes and
31 potential themes. Codes relating to the barriers and facilitators to PA will be mapped onto the TDF.²¹
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22 These codes will then be applied by the researcher to remaining interviews, incorporating any new
23 codes and themes as they are identified. For analytical rigour, the classification of belief statements
24 to the TDF domains will be discussed and agreed within the team. Coding and analysis will be
25 facilitated by QSR International's NVivo software (Version 12, 2018).

61 WP2: Co-design activities

62 Analysis of co-design activities will be focused specifically on the aims of each activity, pragmatic and
63 expeditious so findings can be fed into subsequent stages.³⁷ Qualitative content analysis will be
64 performed using QSR International's NVivo software (Version 12, 2018), supplemented by other
65 forms of analysis as required.
66

67 **Ethics and dissemination**

68 A favourable opinion has been granted from the North East - Tyne & Wear South Research Ethics
69 Committee (REC ref: 18/NE/0274).

70 Informed consent will be sought prior to participation. Participants will be informed that
71 participation is entirely voluntary, and they may withdraw at any point, without giving a reason and
72 without negative consequences. They will be asked for their agreement to audio/video record
73 (where relevant) and informed that recordings are confidential, and transcriptions of audio-

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3 recordings will be anonymised. Ethical considerations relating to the interviewing of children (aged
4 under 16 years) are described above. Interview participants will be offered payment of any travel
5 expenses and a £20 high-street shopping voucher. This amount is based on the need to provide
6 some compensation for the participant's time, expertise and contribution to the research but
7 without coercing individuals to take part when they would rather not.⁴⁰ Participants will be notified
8 of the voucher in the study information sheet and will be offered the voucher at the beginning of the
9 interview to convey to them that they are being rewarded for their attendance, and not for what
10 they share during the interview.⁴¹ Participants recruited to co-design activities will receive a high
11 street voucher which reflects the time commitment and nature of the activity they choose to
12 participate in, in accordance with INVOLVE guidance.⁴²

13
14 Findings will be disseminated via our study website (<https://research.ncl.ac.uk/beingactive/>),
15 conferences and journal publication. A summary of research findings will be available for
16 participants. PPI will inform further dissemination activities (e.g. via patient organisations, social
17 media), appropriate formats (e.g. infographics, video) and content to ensure lay summaries are
18 understandable and engaging to survivors.

21 22 23 24 25 **Patient and public involvement**

26 Feedback on the study concept and methods were gained from two established PPI groups in
27 Newcastle: the Young Person's Advisory Group – North England (YPAG-NE) whose members are
28 young people aged 13-18 years old, and the Perspectives in Cancer Research group whose members
29 are survivors of adult cancer. Views of young adult CCSs were gained via representatives of two
30 European cancer organisations, PanCare (Pan-European Network for Care of Survivors after
31 Childhood and Adolescent Cancer) and Youth Cancer Europe, and a survey posted to a closed
32 Facebook group for survivors of cancer. YPAG-NE and two adult CCSs provided comments on patient
33 information and interview topic guides/cue cards. Patient representatives (OB & JH) sit on the
34 project steering group, and will be involved in data interpretation, co-design activities, and advising
35 on dissemination.

38 39 40 41 **DISCUSSION**

42 A high proportion of adolescents and young adults do not meet recommended levels of PA,⁴³ and
43 there are already many publicly available programmes and interventions to encourage PA, including
44 government/health service initiatives and apps.⁴⁴⁻⁴⁶ This raises the question of whether a specific PA
45 intervention is needed for CCSs. While there is considerable overlap between determinants of PA in
46 CCSs and young people without cancer, many influences are likely specific to CCSs (e.g. cancer-
47 related fatigue, frustration about impact of cancer).¹³ There are also concerns about the quality and
48 likely effectiveness of many of the publicly-available PA programmes.⁴⁶ In addition, cancer survivors
49 may question the relevance of general (i.e. non-cancer specific) PA programmes to them,⁴⁷ and
50 there is evidence that tailoring interventions to a specific target population is likely to increase
51 effectiveness.⁴⁸ Taken together, this suggests that the route most likely to lead to changes in CCSs'
52 PA levels is to develop an intervention specifically for this group.

53
54 The strength of our study lies in the adoption of an evidence-based, person-centred approach.
55 However, we also recognise the need to mitigate potential study limitations. To minimise selection
56 bias, the importance of giving all eligible patients the opportunity to hear about the study, and
57 allowing them make their own choice as to whether they want to participate or not, will be

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3 highlighted to those involved in the screening process.⁴⁹ For example, participating in this research
4 may appeal more to CCSs who are physically active, than to those who are not. Therefore, patients
5 will also be made aware that a judgement will not be made on their current activity levels and that
6 we are interested in their views regardless of whether they consider themselves to be active or not.
7

8
9 Although several determinants of PA may be common across CCSs as a group (e.g. fatigue), survivors
10 will experience barriers and limitations specific to the cancer they had, and the treatment they
11 received.⁵⁰ Many CCSs have ongoing health conditions and impairments to the neurological,
12 endocrine, musculoskeletal and cardiopulmonary systems which can influence physical
13 performance, function and mobility in a variety of ways.⁵¹ Therefore, it is essential that PA
14 interventions should not only be targeted to the needs of CCSs as a group, but allow tailoring to the
15 needs of individuals.¹³ To enable this, recruitment will occur via follow-up clinics for survivors of
16 haematological malignancies, solid tumour and central nervous system tumours, and sampling will
17 ensure variation across key characteristics including diagnosis and treatment to ensure
18 heterogeneity in the potential influences on the PA of CCSs.
19

20
21 CCSs can be challenging to recruit to research,⁵² therefore, we propose several routes by which CCSs
22 may be made aware of the study. This will help to safeguard against any potential recruitment issues
23 and will also ensure that a wide range of individuals are offered the opportunity to participate. We
24 also acknowledge the potential impact of the current COVID-19 pandemic on the study, including
25 the possible need to conduct co-design activities remotely. The use of video-conferencing could
26 exclude those who have limited access to the required technology, or those who do not feel
27 comfortable using it.⁵³ The use of remote methods may also hinder the interactive, creative and
28 collaborative process essential to co-design. Therefore, we have proposed several ways that
29 individuals can take part in co-design activities, including online synchronous and asynchronous
30 methods. Careful and considered planning will be needed to adapt co-design activities to ensure
31 participation and engagement, as well as an online environment in which individuals feel safe and
32 able to contribute.
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35 The final output from this phase of the BEACON study will be a prototype evidence-based and
36 theoretically-informed intervention. The next step will be to fully operationalise the intervention and
37 any supporting materials (e.g. training manual). Efficient and systematic user pre-testing studies will
38 be conducted to provide insight into different aspects of the intervention and iteratively refine and
39 optimise it.³⁷ Subsequently, as recommended in the area of PA research,⁵⁴ we plan to assess
40 feasibility and acceptability to users and, following that, evaluate effectiveness and cost-
41 effectiveness in a randomised controlled trial, with a parallel process evaluation.⁵⁵
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45 Various organisations, including the American Cancer Society⁵⁶ and Macmillan Cancer Support,⁵⁷
46 have produced PA recommendations for cancer survivors (of all ages). However, understanding
47 remains limited on how best to support survivors to improve levels of PA and maintain changes.^{58 59}
48 The study described here – although it focuses on CCSs – provides an example of how to use a
49 behavioural science approach to develop a person-centred, evidence-based and theoretically-
50 informed PA intervention and, therefore, may be informative for those interested in systematically
51 developing PA interventions for other survivor groups.
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54 **Author contributions**

55
56 MB, LS, RS and VAS conceived the study. MB, LS, RS, VAS, NS and AG developed the grant proposal
57 and secured funding. MB managed the ethical and research approvals and will undertake the field
58 work and data analysis supported primarily by LS and VAS. RS, AG and NS will support recruitment of
59 participants. JH & OB have, and will continue to, provide PPI input. JS and KM will provide specialist
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3 advice and expertise throughout the study. MB drafted the manuscript, and AG, JS, LS, RS, KM, NS,
4 VAS provided revisions and comments. All authors approved the final manuscript.
5

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7 number CCLGA 2017 26, North of England Children's Cancer Research, and the Children's Cancer
8 Fund.
9

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12 **Competing interests statement:** No interests to declare.
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16 **Figure 1:** Overview of planned research for the BEACON project
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18 **Table 1:** Domains, actions and planned methods to develop physical activity intervention for
19 childhood cancer survivors
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21 **Table 2:** Childhood cancer survivors' inclusion and exclusion criteria
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Table 1: Domains, actions and planned methods to develop physical activity intervention for childhood cancer survivors

Key actions to consider for intervention development ¹⁶	Domain of intervention development and associated specific action(s) ¹⁵	Methods utilised, or planned in this research to develop intervention
Planning the process Involving stakeholders Bringing together a team	Conception	
	1. Identifying the problem in need of a new intervention (including the health problem, the problematic behaviour and the target population)	<ul style="list-style-type: none"> • Identification and evaluation of the literature on: <ul style="list-style-type: none"> - Prevalence of cardiovascular late effects in CCSs - Low PA in CCSs - Benefit of PA to the health of CCSs • Clinical experience/knowledge of low PA in CCSs.
Reviewing published evidence Undertaking primary data collection Drawing on existing theories Articulate programme theory Understanding context Attending to future implementation	Planning	
	2. Setting up a planning group/development team	<ul style="list-style-type: none"> • Establishing a multidisciplinary steering group involving: <ul style="list-style-type: none"> - Researchers and academics with expertise in health psychology, behavioural science/intervention development, exercise physiology, PA interventions in clinical populations, digital health innovation. - HCPs and service providers (consultant oncologists, nurse specialists). - Patient representatives.
	3. Understanding the problem to be addressed <i>i. Understanding the views and experiences and psycho-social of the potential target population</i>	<ul style="list-style-type: none"> • Use of patient and public involvement during initial stages of planning: <ul style="list-style-type: none"> - Gaining young peoples' views of PA and the study concept via an NHS young persons' advisory group - Consultation with CCSs on the need for PA interventions via focus group and survey methods • Identification of literature reporting HCPs (who may be involved in the resulting intervention) views of PA in CCSs, and the provision of PA advice to CCSs.
	<i>ii. Assessing the causes of the problems</i>	<ul style="list-style-type: none"> • Undertaking research to explore and understand CCSs views and experiences of PA <ul style="list-style-type: none"> - Literature review of the barriers and facilitators to PA in CCSs - Undertaking in-depth interviews with CCSs and their parents regarding their views and experiences of PA, including perceived barriers and facilitators (informed by the Theoretical Domains Framework) - Creating a logic model of the problem
<i>iii. Describing and understanding the wider context of the target population and the context in which the intervention will be implemented</i>	<ul style="list-style-type: none"> • Undertaking research to explore the views of CCSs views of receiving lifestyle/PA advice in follow-up care. <ul style="list-style-type: none"> - Survey and interviews with CCSs attending follow-up care • Involving HCPs, service providers and patients in steering group and in co-production of intervention. 	
<i>iv. Identifying the effectiveness of interventions for PA in CCSs</i>	<ul style="list-style-type: none"> • Identification and evaluation of existing PA interventions in CCSs. <ul style="list-style-type: none"> - Systematic reviews of PA interventions in CCSs - Research evaluating PA interventions in CCSs 	

	v. <i>Understanding wider stakeholders' perspectives of problems and issues</i>	<ul style="list-style-type: none"> Actively engage with stakeholders, service providers and patients throughout research and in co-production of intervention.
Drawing on existing theories Articulating programme theory Understanding context Attending to future implementation	4. <i>Making decisions about aims and goals of intervention</i>	Based on the evidence generated in WP1, the steering group and research team will make decisions on the specific aims and goals of intervention. These will be presented and discussed in co-design workshops.
	5. <i>Identifying what needs to change, how to bring about change</i>	Following WP1 a logical model of change will be developed for PA in CCS drawing on the evidence and constructs from relevant theories.
	6. <i>Specify who will change, how and when</i>	<ul style="list-style-type: none"> Following WP1 the steering group and research team will break down the behavioural outcomes to consider, prioritise and map who needs to change what, how changes will occur as a result of the intervention and when these changes are expected to take place.
	7. <i>Considering the real-world issues about cost and delivery of any intervention to reduce risk of implementation failure</i>	<ul style="list-style-type: none"> Involvement of HCPs/service providers in development team Co-design workshops with stakeholders (including HCPs) Use of Normalisation Theory Process to inform discussion with HCPs
	8. <i>Considering whether it is worthwhile continuing with development of intervention</i>	<ul style="list-style-type: none"> Steering group and stakeholder input on feasibility of intervention
Designing and refining intervention	Designing	
	9. <i>Generating ideas about solutions and components and features of an intervention</i>	<ul style="list-style-type: none"> Mapping of behavioural determinants onto behaviour change techniques using Behaviour Change Wheel. Co-design workshops using creative methods and activities to enable idea generation
	10. <i>Re-visit decisions about where to intervene</i>	<ul style="list-style-type: none"> Input from stakeholders to make final decisions regarding the scope, the target population, key features and components of intervention which will be further refined during workshops.
	11. <i>Make decisions about the content, format and delivery of the intervention</i>	<ul style="list-style-type: none"> Findings of WP1 will be combined into a theoretical model of PA in CCSs and to develop initial ideas about content, format and delivery. Actively engaging with steering group and stakeholders via co-design workshops to obtain views on the potential content, format and delivery of intervention
	12. <i>Design an implementation plan, thinking about who will adopt the intervention and maintain it</i>	<ul style="list-style-type: none"> Design of potential implementation plan will be informed by discussions with HCPs and other stakeholders regarding potential implementation barriers and previous research.
	Creating	
	13. <i>Make prototypes/mock-ups of the intervention where relevant</i>	<ul style="list-style-type: none"> Generation and discussion of mock-ups and paper-based prototypes during co-design workshop.

Table 2: Childhood cancer survivors' inclusion and exclusion criteria

Inclusion criteria	Exclusion criteria
Diagnosed with any haematological malignancy or solid or CNS tumour under the age of 19 years	Any cognitive or physical impairment of sufficient severity to limit their ability to understand, engage with or undertake PA
Currently aged 10-24 years	Any contraindications to exercise
Currently 2-15 years from the end of treatment	
No active disease	

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Input from steering group and PPI

Workpackage 1

In-depth interviews informed by theory

To identify perceived barriers and facilitators to PA in CCSs

CCSs recruited from two NHS trusts

Parents of CCSs recruited from two NHS trusts

Thematic analysis of interview data

Previous primary research with CSSs and systematic reviews of relevant literature will also inform work package 2

Workpackage 2

Phase 1: Behavioural analysis

Identifying potentially modifiable determinants of PA in CCSs and developing a theoretical model of PA in CCSs. Identifying potential intervention functions, components and behaviour change techniques

Phase 2: Co-design process

Stage 1 co-design activities
Testing evidence statements
Generate new ideas for interventions

Participants:
CCSs, parents, HCPs, academics/researchers

Output:
Intervention principles

Stage 2 co-design activities
User feedback on intervention principles
Develop core intervention ideas/concepts
Design prototypes of potential interventions

Participants:
CCSs, parents, HCPs, academics/researchers

Output:
Core concepts
Paper prototypes

Final output: Prototype intervention

Childhood cancer survivors (CCSs); Healthcare professionals (HCPs); Physical activity (PA)

Figure 1: Overview of planned research for the BEACON project