Heeding the psychological concerns of young cancer survivors: A single-arm feasibility trial of CBT and a cognitive behavioral conceptualization of distress

Josefin Hagström [1], **Malin Ander** [1], **Martin Cernvall** [1], **Brjánn Ljótsson** [2], **Henrik W Wiman** [1], **Louise von Essen** [1], **Joanne Woodford** [Corresp. 1]

[1] Clinical Psychology in Healthcare, Department of Women's and Children's Health, Uppsala Universitet, Uppsala, Sweden

[2] Division of Psychology, Department of Clinical Neuroscience, Karolinska Institute, Stockholm, Sweden

Corresponding Author: Joanne Woodford
Email address: joanne.woodford@kbh.uu.se

Background: A subgroup of adolescent and young adult (AYA) survivors of cancer during adolescence report high levels of psychological distress. To date, evidence-based psychological interventions tailored to the cancer-related concerns experienced by this population are lacking. The present study aimed to (1) examine the feasibility and preliminary efficacy of an individualized cognitive behavioral therapy (CBT) intervention for AYA survivors of cancer during adolescence; and (2) identify and conceptualize cancer-related concerns as well as maintaining factors using cognitive-behavioral theory.

Methods: A single-arm trial, whereby AYA survivors of cancer during adolescence (aged 17-25 years) were provided individualized face-to-face CBT at a maximum of 15 sessions. Clinical outcomes were assessed at baseline, post-intervention, and three-month follow-up. Intervention uptake, retention, intervention delivery, and reliable change index scores were examined. An embedded qualitative study consisted of two unstructured interviews with each participant pre-intervention. Along with individual behavioral case formulations developed to guide the intervention, interview data was analyzed to identify and conceptualize cancer-related concerns and potential maintaining factors.

Results: Ten out of 213 potential participants invited into the study was included, resulting in an overall participation rate of 4.7%. Nine participants completed the intervention, with respectively seven and eight participants completing the post-intervention and three month follow-up assessment. The majority of reported cancer-related concerns and maintaining factors were conceptualized into four themes: social avoidance, fear of emotions and bodily symptoms, imbalance in activity, and worry and rumination.

Conclusions: Given significant recruitment difficulties, further research is required to examine barriers to help-seeking in the AYA cancer survivor population. However, the conceptualization of cancer-related concerns and maintaining factors experienced by the population may represent an important first step in the development of psychological support tailored toward AYA cancer survivors' unique needs.

Heeding the psychological concerns of young cancer survivors: A single-arm feasibility trial of CBT and a cognitive behavioral conceptualization of distress

Authors

Josefin Hagström[1], MSc, Malin Ander[1], PhD, Martin Cernvall[1], PhD, Brjánn Ljótsson[2], PhD, Henrik W. Wiman[1], MSc, Louise von Essen[1], PhD, Joanne Woodford[1†], PhD

Affiliations

[1] Clinical Psychology in Healthcare, Department of Women's and Children's Health, Uppsala Universitet, Uppsala, Sweden

[2] Division of Psychology, Department of Clinical Neuroscience, Karolinska Institute, Stockholm, Sweden

Corresponding Author

†Joanne Woodford

E-mail: joanne.woodford@kbh.uu.se

Abstract

Background: A subgroup of adolescent and young adult (AYA) survivors of cancer during adolescence report high levels of psychological distress. To date, evidence-based psychological interventions tailored to the cancer-related concerns experienced by this population are lacking. The present study aimed to (1) examine the feasibility and preliminary efficacy of an individualized cognitive behavioral therapy (CBT) intervention for AYA survivors of cancer during adolescence; and (2) identify and conceptualize cancer-related concerns as well as maintaining factors using cognitive-behavioral theory.

Methods: A single-arm trial, whereby AYA survivors of cancer during adolescence (aged 17-25 years) were provided individualized face-to-face CBT at a maximum of 15 sessions. Clinical outcomes were assessed at baseline, post-intervention, and three-month follow-up. Intervention uptake, retention, intervention delivery, and reliable change index scores were examined. An embedded qualitative study consisted of two unstructured interviews with each participant pre-intervention. Along with individual behavioral case formulations developed to guide the intervention, interview data was analyzed to identify and conceptualize cancer-related concerns and potential maintaining factors.

Results: Ten out of 213 potential participants invited into the study was included, resulting in an overall participation rate of 4.7%. Nine participants completed the intervention, with respectively seven and eight participants completing the post-intervention and three month follow-up assessment. The majority of reported cancer-related concerns and maintaining factors were conceptualized into four themes: social avoidance, fear of emotions and bodily symptoms, imbalance in activity, and worry and rumination.

Conclusions: Given significant recruitment difficulties, further research is required to examine barriers to help-seeking in the AYA cancer survivor population. However, the conceptualization of cancer-related concerns and maintaining factors experienced by the population may represent an important first step in the development of psychological support tailored toward AYA cancer survivors' unique needs.

Introduction

Adolescent and young adult (AYA) survivors of cancer diagnosed during adolescence report numerous stressors relating to cancer diagnosis, treatment, and survivorship (Abrams, Hazen, & Penson, 2007; Lehmann et al., 2014). The adolescent period includes challenges in terms of development as well as transition into adulthood (Jaworska & MacQueen, 2015), and the onset of several common mental health difficulties (Kessler et al., 2012). As such, AYAs diagnosed with cancer during adolescence appear at an increased risk of problems related to their mental health. Indeed, studies have demonstrated that AYA survivors of cancer during adolescence report elevated levels of psychological distress in comparison to matched cancer-free controls (Seitz et al., 2010), those diagnosed during childhood (Kazak et al., 2010) and adulthood respectively (Lang et al., 2018). Further, research indicates that receiving a cancer diagnosis prior to age 25 leads to a higher risk of suicide (Gunnes et al., 2017) and being prescribed anxiolytics and hypnotics (Johannsdottir et al., 2018). Although some evidence has suggested lower levels of psychological distress compared to population norms (Larsson, Mattsson, & von Essen, 2010), research has consistently identified a subgroup of AYA survivors of cancer during adolescence who report an elevated level of long-term psychological distress (de Laage et al., 2016; Husson et al., 2017).

Despite a significant subgroup of AYA survivors of cancer during adolescence reporting an elevated level of psychological distress, the group reports unmet needs of psychological support (Bibby et al., 2017; Kaul et al., 2017). As a result, the development of psychological interventions adapted for the unique concerns of this group has been posited (Ander et al., 2016; Seitz, Besier, & Goldbeck, 2009; Seitz et al., 2010). One potential solution may be the provision of psychological support based on cognitive-behavioral therapy (CBT), an evidence-based approach that has been demonstrated to be effective for a range of mental health difficulties (Kaczkurkin & Foa, 2015; McMain et al., 2015). Indeed, CBT-based interventions have shown promise in reducing psychological distress in AYAs diagnosed with cancer during childhood (Fisher et al., 2015; Seitz et al., 2014; van der Gucht et al., 2017). Whilst some recognition has been given the specific and unique challenges associated with being diagnosed with cancer during adolescence (Abrams, Hazen & Penson, 2007; Seitz, Besier, & Goldbeck, 2009), so far only a few CBT based psychological interventions have been developed with adaptation and tailoring to this population (Campo et al., 2017; Sansom-Daly et al., 2018).

In order to overcome this psychological treatment gap, a thorough theoretical understanding is needed concerning symptoms of psychological distress and mechanisms involved in the maintenance of distress (Craig et al., 2008). Two models have been proposed to guide the development of psychological interventions for adolescents diagnosed with cancer: (1) the Adolescence Resilience Model (Haase, 2004) and (2) the Pediatric Medical Traumatic Stress Model (Kazak et al., 2006). However, neither model recognizes the full range of areas of cancer-related distress experienced by AYA survivors of cancer during adolescence, such as feelings of

worthlessness and feeling left behind (Ander et al., 2018). Nor does either model specify the concerns or mechanisms hypothesized to be involved in the maintenance of distress. Further, psychological interventions based on these models have shown limited effect on the reduction of cancer-related distress (Kazak et al., 2004; Robb et al., 2014). Research to identify mechanisms involved in the maintenance of distress has started (Sansom-Daly & Wakefield, 2013), however there is a need to further extend the knowledge of the different manifestations of psychological distress experienced by the population, alongside an examination of cancer-related concerns and maintaining factors for distress. Such knowledge can be used to guide development of psychological interventions tailored for AYA survivors of cancer during adolescence (Seitz et al., 2010).

Seeking to inform the development of a CBT-based intervention tailored for AYA survivors of cancer during adolescence, the present study had a twofold primary aim: (1) to examine the feasibility (e.g., recruitment, attrition, data collection, intervention procedures) and preliminary efficacy of an individualized CBT intervention for the population; and (2) to develop a cognitive-behavioral theory-based conceptualization of cancer-related psychological concerns and maintaining factors experienced by the population.

Materials & Methods

Study design

A single-arm trial design with a pre-post and three month follow-up design, and embedded qualitative study, was adopted to examine the feasibility and preliminary efficacy of individualized CBT and develop a conceptualization of cancer-related psychological distress. The study was approved by the Uppsala Regional Ethical Review Board (reference number: 2014/443).

Setting

Delivery of individualized CBT and data collection was undertaken at two locations: the pediatric oncology unit at the Children's University Hospital in Uppsala and a private psychology practice in Stockholm.

Participants

Participants were eligible if: (1) aged between 15-25 years; (2) diagnosed with cancer during adolescence (13-19 years); (3) completed cancer treatment at the pediatric oncology center in Uppsala or Stockholm (according to pediatric oncology centers for persons 15-17 years, or self-report for persons ≥18 years); (4) able to participate in individual CBT once a week for up to 15 weeks in Uppsala or Stockholm; and (5) a self-reported need for psychological support. Potential participants were excluded if: receiving a psychological intervention; and/or a self-reported

severe and/or enduring mental health problem (e.g. bipolar disorder, psychosis) and/or acute suicidality requiring more specialized treatment than could be offered within the present study.

Recruitment and study procedures

Recruitment took place from February through October 2015. First, potential participants diagnosed with cancer during adolescence, meeting the inclusion criteria, were identified via the Swedish Childhood Cancer Registry. Second, their telephone numbers were retrieved either via internet search engines (18-25 years) or pediatric oncology centers (15-17 years). All potential participants, whereby a telephone number could be identified, were telephoned by one of the research team members in two waves: first potential participants treated in Uppsala; and thereafter potential participants treated in Stockholm. In cases whereby the individual could not be invited via telephone a study invitation letter was sent via post, including a link to a study website where one could register contact details if interested in participating. Study invitation letters informed potential participants that the study was to examine CBT for AYA survivors of cancer during adolescence who currently experienced psychological difficulties related to the cancer experience. Potential participants were informed that the intervention would last 10-15 weeks and that therapeutic work would focus on helping them with difficult thoughts, feelings and behaviors.

All potential participants expressing interest to participate were invited to a face-to-face screening assessment with a study therapist (two licensed psychologists and one psychologist in clinical training). The Montgomery Åsberg Depression Rating Scale-Self Assessment (MADRS-S) (Svanborg & Åsberg, 1994) and Mini International Neuropsychiatric Interview (M.I.N.I.) version 5.0 (Sheehan et al., 1998) were administered to screen for depression and anxiety disorders (including PTSD) and to identify individuals with a severe or enduring mental health problem (e.g. bipolar disorder, psychosis) and/or acute suicidality. Individuals with such difficulties were excluded and guided to appropriate support. For one participant who was under the age of 18 at the study start, parents were provided with written information and informed about the study via telephone. In addition, an assessment was conducted of the participant's understanding of the study to determine if there was a need for parental consent.

Written information about study participation was provided to potential participants prior to the screening assessment. If found eligible, study participation was offered and written informed consent obtained. Subsequently, baseline assessments, and two unstructured interviews were conducted. The screening, baseline assessments, and interviews took place over one-to-three sessions. After completion of the baseline assessments, participants were assigned to one of three study therapists. Two of the three study therapists were co-authors (MA and MC). In eight cases, the same study therapist who conducted the screening and baseline assessment also conducted subsequent interviews and delivered the intervention. Except for one case, post- and follow-up assessments were conducted by another study therapist than the one providing the intervention.

164
165 **Sample size**
166 As a primary aim was to examine feasibility (e.g., recruitment, data collection, intervention
167 delivery), a formal sample size calculation was not conducted (Billingham, Whitehead & Julious,
168 2013). Study recruitment was stopped when all individuals identified via permission by the
169 ethical committee had been screened for eligibility. Few psychological intervention studies have
170 been conducted among AYA survivors of cancer during adolescence (Barnett et al., 2016) and
171 estimations of recruitment rates were difficult to determine a priori. However, as informed by
172 large mental health intervention trials in the general population, a recruitment rate of 12% may
173 have been anticipated (Richards et al., 2013; Richards et al., 2016). At the time of the trial, 213
174 potentially eligible AYA cancer survivors were registered in the Swedish Childhood Cancer
175 Registry in the Stockholm and Uppsala areas. Taking the estimate of a 12% recruitment rate, a
176 sample size of 26 had been anticipated.
177
178 **Data collection**
179 *Sociodemographics:* Data for the following variables was collected via the Swedish Childhood
180 Cancer Registry: (1) current age; (2) age at diagnosis; (3) gender; and (4) cancer diagnosis. Self-
181 reported information at baseline assessment included: (1) cancer recurrence; (2) time since end
182 of cancer treatment; (3) type of cancer treatment; (4) number and type of late effects; (5)
183 previous receipt of psychological intervention; (6) accommodation status; (7) relationship status;
184 and (8) employment status.
185
186 *Feasibility outcomes:* Feasibility outcomes related to recruitment, attrition, data collection, and
187 intervention procedures were collected.
188
189 *Clinical outcomes:* A number of self-report clinical outcome measurements (Swedish
190 translations) were administered at baseline, post-intervention, and three-month follow-up. The
191 MADRS-S and the M.I.N.I. version 5.0 were re-administered at post-intervention and three-
192 month follow-up.
193
194 Symptoms of anxiety were assessed using the 21-item Beck Anxiety Inventory (BAI) (Beck et
195 al., 1988). Posttraumatic stress symptoms were assessed using the 17-item PTSD Checklist –
196 Civilian Version (PCL-C) (Weathers et al., 1993) as defined in the B (re-experiencing), C
197 (avoidance), and D (hyperactivity) criteria in DSM-IV (American Psychiatric Association,
198 2000), with items modified to focus on the participant's experience of cancer. The 16-item Penn
199 State Worry Questionnaire (PSWQ) (Meyer et al., 1990) was used to assess worry, and the 10-
200 item Body Image Scale (BIS) (Hopwood et al., 2001) to measure body dissatisfaction. Health
201 anxiety was assessed using the 18-item Short Health Anxiety Inventory (SHAI) (Salkovskis et
202 al., 2002) and the Patient Health Questionnaire-15 (PHQ-15) (Kroenke, Spitzer & Williams,
203 2002) was adopted to measure somatic symptoms. The 10-item Fatigue Assessment Scale (FAS)
204 (Michielsen, de Vries & van Heck, 2003) was used to assess fatigue. Rumination was assessed

using the 22-item Rumination Scale of the Response Style Questionnaire (RSQ) (Nolen-Hoeksema, 1991) and experiential avoidance was examined using the 10-item version of the Acceptance and Action Questionnaire (AAQ-II) (Bond et al., 2011). Functional impairment in work, social, and family life was measured using the three-item Sheehan Disability Scale (SDS) (Sheehan, 1983).

Unstructured interviews: Prior to commencing the intervention, two unstructured interviews (lasting 33 to 104 minutes) were conducted with each participant over a two-week period by the therapist the participant had been assigned to. All interviews began with the question: "*Please tell me how you think and feel about having had cancer?*." Subsequently, probes such as "*Tell me more about…*", "*Tell me how you felt when…*", and "*Please tell me what you mean by…*" were used to encourage the participant to provide more in depth responses and facilitate clarification (Ander et al., 2018). Before the second interview, the interviewer listened to a recording of the first interview to identify areas for further exploration. To improve interview quality, a senior researcher not otherwise associated with the study, with extensive experience in qualitative interviewing, reviewed one interview per therapist.

Intervention

The intervention was delivered by two licensed psychologists and one psychologist in clinical training across the two sites. Given the explorative nature of the study, cognitive-behavioral case formulations were developed by study therapists, including the identification of current and previous problems and concerns, prioritization of concerns and specification of topography (i.e. descriptions of the specific behavioral features). The subsequent intervention was tailored to each participant's individual problems and needs according to these case formulations (Persons, 2008). Further, functional analyses were developed using learning theory, to make explanatory inferences partly on the association between precipitating events and resulting problems, and partly on factors hypothesized to have maintained individuals' problems (Dougher, 2000; Sturmey, 2008).

An initial working cognitive-behavioral case formulation was developed over intervention sessions one-to-three, and subsequently tested and refined during the course of the intervention. Study therapists used relevant cognitive-behavioral models and standard CBT techniques dependent on the main problems raised by participants. CBT techniques were discussed among study therapists during continuous supervision however individual study therapists made final decisions concerning the selection of CBT techniques. An overview of delivered techniques can be seen in Table 1.

Table 1:
Number of participants receiving the respective CBT techniques.

INSERT TABLE 1 ABOUT HERE

Each participant was offered a maximum of 15 sessions of individual CBT (each session lasting approximately 45 minutes). Cognitive-behavioral case formulations was discussed with the other study therapists, in supervision by a licensed psychologist with expertise in delivering CBT to young people experiencing mental health problems and clinical behavior analysis, and in external supervision of a licensed psychologist with expertise in clinical behavior analysis. Both supervisors were external to the study team. Formulations were also discussed during internal supervision to further receive opinions and increase the validity of the formulations

Data analysis

An adapted version of the Consolidated Standards of Reporting Trials (CONSORT) diagram for parallel randomized trials (Moher et al., 2010) was used to illustrate participant flow. Descriptive statistics (means and SDs) were used to report clinical outcomes at baseline, post-treatment, and three-month follow-up assessments. Descriptive statistics were used to report feasibility outcomes pertaining to recruitment, attrition, data collection, and treatment delivery. As a preliminary investigation of efficacy, the proportion of participants reporting a reliable change in scores on the MADRS-S, BAI, and/or PCL-C was calculated according to the Jacobson-Truax Index (Jacobson & Truax, 1991), with test-retest reliability of the measures from previous studies obtained (Fydrich, Dowdall & Chambless, 1992; Ruggiero et al., 2003; Svanborg & Åsberg, 1994) to inform the calculation. Missing items were imputed using the mean item score of available items on the outcome measurement in cases whereby a maximum of two, or 10%, of items in a measurement, were missing. List-wise deletion was used for the calculation of RCI for the cases whereby baseline-, post-, or three month follow-up assessment was missing. As such, participants were excluded from analysis if their post-intervention and/or three-month follow-up assessment was missing.

No formal method of analyzing the qualitative interview data was adopted. However, the method was informed by steps utilized in qualitative framework analysis (Ritchie & Spencer, 1994) and theoretical thematic analysis (Braun & Clarke, 2006). A framework was developed a priori for extracting and summarizing the data for the purpose of categorizing cancer-related and non-cancer-related concerns and key characteristics informed by CBT (e.g., current and previous concerns, topography, maintaining factors). Moreover, informal discussions and reflections regarding the conceptualization and emerging themes were ongoing among therapists throughout the study.

First, with the aim of identifying current and previous cancer-related concerns alongside, topography and maintaining factors, interview transcripts, clinical notes, and behavioral case formulations were read by the second author (MA) to gain a sense of the dataset. Behavioral case formulations were thereafter read by MA to identify cancer-related concerns which were refined

into themes and defining key characteristics informed by CBT. Thereafter, interview transcripts were re-read by MA to extract cancer-related concerns considered relevant for each theme. The third author (MC) reviewed the themes of cancer-related concerns and experiences with respect to coherence, congruence, and distinctiveness (Braun & Clarke, 2006) and themes were subsequently elaborated and refined. CBT case formulations and clinical notes were further examined by MA, to identify and summarize the specific CBT techniques applied during the intervention (see Table 1). Themes were finally reviewed, alongside the original data by MC to further increase trustworthiness.

Results

Demographic and clinical outcomes

Participant characteristics are provided in Table 2. According to the cancer registry, the majority suffered from leukemia (50%). Most reported living with their parents (60%); no history of relapse (90%); having experienced late effects (80%); and previous receipt of psychological support at some point in life (69%). One participant self-reported being diagnosed with Myelodysplastic syndrome (MDS) which contrasted with registry information.

Table 2:
Participant characteristics (*N*=10).

INSERT TABLE 2 ABOUT HERE

Means, standard deviations for clinical outcomes at baseline, post-treatment, and three-month follow-up assessment are summarized in Table 3.

Table 3:
Means, standard deviations, and ranges for the clinical outcomes at baseline, post-intervention, and three month follow-up assessment (*N*=10).

INSERT TABLE 3 ABOUT HERE

The proportions of participants meeting criteria for reliable change on the MADRS-S, BAI, and PCL-C were calculated for seven participants providing complete outcome data at post-intervention and three-month follow-up and are presented in Table 4. At post-intervention, 3/7 (43%) participants met criteria for reliable change on at least one outcome, with 4/7 (57%) meeting criteria at three-month follow-up. Except for one participant's score on the PCL-C, all participants reported reduced scores at follow-up compared to baseline on the self-report measures.

Table 4:
Number of individuals with complete data reaching reliable change from baseline to post-intervention and from baseline to follow-up assessment (*n*=7).

INSERT TABLE 4 ABOUT HERE

Feasibility outcomes

A summary of feasibility outcomes is shown in Table 5, with more detailed data concerning the main feasibility objectives reported below.

Table 5:

Summary of feasibility outcomes.

INSERT TABLE 5 ABOUT HERE

Recruitment: A CONSORT diagram, adapted for a single-armed trial, illustrating the flow of participants through the study, including reasons for non-participation, is shown in Figure 1.

INSERT FIGURE 1 ABOUT HERE

Figure 1: Adapted CONSORT diagram illustrating the flow of participants through the study.

Out of 286 initially identified individuals via the Swedish Childhood Cancer Registry, 213 potential participants (213/286; 75%) were to be contacted, with the remaining 73 individuals excluded (see Figure 1 for reasons). Of those, 209 were contacted and received study information via telephone (111/209; 53%), or the post (98/209; 47%). Nine of those invited via telephone (9/111; 8%) and four of those invited via the post (4/98; 4%) expressed interest in study participation and were assessed for eligibility (in total: 13/213, 6%). Of those assessed for eligibility, three were excluded (3/13; 23%) while ten (10/13; 77%) were eligible and allocated to the intervention, resulting in an overall participation rate of 5% (10/213).

Attrition: Out of the ten participants allocated to the intervention, one participant discontinued participation before receipt of the intervention, resulting in a total of nine participants (9/10; 90%) receiving the intervention. Completion rates of assessments were as follows: baseline assessment, n=9; post-intervention assessment, n=7; and three month follow-up assessment, n=8.

Data collection: The last post-intervention assessment was conducted in April 2016 and the last three month follow-up assessment in June 2016. The mean time between post-intervention and follow-up assessment was 12 weeks (ranging from 10 to 14 weeks). In total, nine items were missing from the self-report questionnaires (one in PHQ-15, two in SHAI-Reassurance subscale at baseline; two in BAI, one in BIS and one in PHQ-15 at post-intervention; one in AAQ-II and one in PHQ-15 at follow-up). Given the low rates of missing items, missing items were imputed using the mean item score of available items on the respective questionnaire for that participant at that specific assessment.

Intervention delivery: Number of intervention sessions ranged from 7 to 15 (mean=13 sessions, median=14 sessions) with a mean intervention length of 25 weeks (range=18-36 weeks), including summer and winter holidays and cancelled intervention sessions. In two cases, the intervention was terminated early despite participants still reporting distress. In one case, the study therapist was unable to complete the intervention and the participant did not want a new study therapist. In the second case, the intervention was terminated due to poor session attendance combined with an identified need for more specialized intervention. The number of cancelled intervention sessions per participant ranged from 0 to 11 (mean=3.4 sessions, median=2 sessions).

Cognitive behavioral conceptualization of cancer-related concerns

A conceptualization was developed, encompassing four themes of cancer-related concerns: social avoidance (n=7), fear of emotions and bodily symptoms (n=7), imbalance in activity (n=8), and worry and rumination (n=8) (the latter was to some extent included within the other themes, yet is presented separately due to being a salient target in the intervention delivered). Each theme is presented in Figure 2 along with defining key characteristics and examples of cancer-related concerns and maintaining factors.

INSERT FIGURE 2 ABOUT HERE

Figure 2: Figure showing identified precipitatory and current cancer-related concerns and maintaining factors.

In addition, a de-identified worked example of an individual's case formulation can be seen in Figure 3, demonstrating how cancer related concerns and maintaining factors operated to maintain problems.

INSERT FIGURE 3 ABOUT HERE

Discussion

To the best of our knowledge, this study represents the first single-arm feasibility study of individualized face-to-face CBT for AYA survivors of cancer during adolescence and conceptualization of cancer-related concerns reported by the population. The conceptualization is consistent with existing models aimed to guide psychological support for AYA cancer survivors (Haase, 2004; Kazak et al., 2006). Importantly, study findings provide a more comprehensive understanding of the distress experienced by the population than existing models, by integrating data from different sources (e.g., case formulations, clinical notes). As such, study results have important clinical implications for further developing psychological support for the population and expand the growing literature on CBT-based interventions for the population (Campo et al., 2017; Sansom-Daly et al., 2018). However, it should be noted that only 10 participants were

recruited raising concerns regarding the feasibility and acceptability of individualized CBT for the population.

Recruitment

Only around 5% of potential participants were included. This finding is in line with research indicating difficulties in recruiting AYA cancer survivors into clinical research (Roth et al., 2016), including trials of psychological interventions (Rabin, Horowitz & Marcus, 2013). Given such recurrent complications, factors that may account for low study inclusion rates should be considered and investigated in future research.

Practical barriers to access: A potential explanation for poor recruitment may pertain to practical barriers in accessing support, as the AYA population has a tendency to relocate relatively frequently as well as being occupied with work, school or extracurricular activities. Indeed, cited reasons for non-participation included travel distance and lack of time. Such findings are consistent with barriers identified by previous work examining psychological interventions for AYA cancer survivors, including burden of travel to attend intervention sessions, time limitations, and scheduling difficulties (Kazak et al., 2004, Barnett et al., 2016). Another notable finding was that 98 (47%) of potential participants were provided with study information via post, as opposed to the telephone, due to incorrect or non-identifiable contact details. This concurs with earlier studies on adult survivors of childhood cancer observing recruitment difficulties due to incorrect contact information (Kilsdonk et al., 2015). As many AYAs undergo transitions in housing, employment, and education, which may cause potential changes in preferred contact methods (Barnett et al., 2016), a wider approach including social media and internet-based methods may be more appropriate. This applies to mode of participant communication as well as the initial identification of potential participants, contrary to merely utilizing cancer registries and centers (Gorman et al., 2014). Indeed, social media and internet-based recruitment strategies have been found to be effective in recruitment of cancer (McCusker et al., 2018) and young adult (Musiat et al., 2016) populations. Whilst in-person recruitment (e.g., in-person recruitment at oncology clinics) has been demonstrated as a successful recruitment strategy in comparison to mail-out, telephone-based, and social media recruitment (Rabin, Horowitz & Marcus, 2013), the necessity of approval for site-specific recruitment can cause delays and negatively affect recruitment (Sansom-Daly et al., 2017). In addition, participant's lack of ability to attend face-to-face therapy may be a reason to explore the potential of internet-administered interventions (Devine et al., 2018; Moody et al., 2015). Offering internet-administered interventions may help overcome some practical barriers to psychological support present in the AYA group.

Helping-seeking behavior: The low participation rate may be explained by findings suggesting that few AYAs show readiness to seek help for psychological distress (Tanielian et al., 2009). Further research suggests that distressed young people struggle to identify, describe, and manage

their emotions (Rickwood et al., 2005). Thus, AYAs may not recognize symptoms of psychological distress (Kovandžić et al., 2011). A further barrier to help-seeking is stigma associated with psychological distress and seeking professional help (Clement et al., 2015), as well as viewing the need for psychological support as a sign of weakness (Salaheddin & Mason, 2016). Indeed, difficulties related to embarrassment and low levels of mental health literacy are commonly reported barriers to seeking support among young adults (Gulliver, Griffiths, & Christensen, 2010). Given struggles with pity and alienation due to the cancer disease (Lehmann et al., 2014), AYA survivors may be reluctant to identify themselves with the additional stigma related to psychological distress. Internet-administered CBT may represent a solution given its potential for increased anonymity and privacy (Younes et al., 2015) potentially overcoming stigma-related barriers. However, research suggests online solutions are commonly designed to fit available technology as opposed to the needs and preferences of end-users (Hollis et al., 2015). As such, to further increase acceptability and engagement, development of an internet-administered CBT intervention should be informed by representatives of the AYA cancer survivor population to ensure needs and preferences are met.

Cognitive behavioral conceptualization of cancer-related concerns

The novel conceptualization of cancer-related concerns stemming from behavioral case formulations and unstructured interviews extends our knowledge in several areas: cancer-related concerns, and maintaining factors of distress within the population. Present findings point to social avoidance, fear of emotions and bodily symptoms, imbalance in activity, and worry and rumination as significant difficulties to address in psychological interventions for AYA survivors of cancer during adolescence.

Social avoidance

One central implication of clinical significance concerns the social difficulties frequently reported by participants, which may seem unsurprising given that social anxiety is common during late adolescence and young adulthood (Gregory et al., 2007). Further, impaired social functioning (Warner et al., 2016; Wilford et al., 2017) and low levels of social participation (Breuer et al., 2017) have been identified as difficulties among AYA cancer survivors. However, there is to-date little research concerning social participation amongst the population, leaving fear of rejection from peers and avoidance of social situations largely unexplored. Additionally, there is currently no evidence-based psychological intervention for AYA survivors of cancer during adolescence specifically targeting social anxiety. Our findings suggest cancer-related experiences of not feeling liked or accepted by peers, fear of causing discomfort and being a burden, and fear of rejection/exclusion, resulting in the adoption of avoidant coping strategies such as post-mortem rumination, refrain from expressing feelings and needs, and social withdrawal. Such concerns and subsequent behaviors may be vital in understanding the social difficulties experienced by the population. Indeed, targeting social skills deficits among young people experiencing social anxiety have been stressed (Mesa, Le & Beidel, 2015) and appear

particularly crucial when working with AYA survivors of brain tumors since the neurocognitive late effects can negatively impact social functioning (Schulte, 2015).

Fear of emotions and bodily symptoms

A second clinical implication for AYA survivors of cancer is based on participants' reports of health anxiety and refers to a fear of emotions and bodily symptoms, such as intrusive cancer-related thoughts and memories, loneliness and health anxiety, resulting in maintaining factors including avoidance behaviors and obsessions and compulsions. The findings are consistent with literature demonstrating increases in anxiety sensitivity (e.g., fear of anxiety symptoms including body sensations) in adolescents who have experienced stressful life events involving serious illness or death (McLaughlin & Hatzenbuehler, 2009). In addition, fear of cancer recurrence has been found to be higher in AYA cancer survivors than older (Cho & Park, 2017) and mixed-age survivors (Thewes et al., 2017). Further, exposure to health-related stressors, combined with low competence in emotion regulation in fear situations observed in adolescents (Zimmermann & Iwanski, 2014) may increase experiential avoidance (Hayes et al., 1996), which predicts the onset and maintenance of anxiety disorders (Spinhoven, van Hemert & Penninx, 2017). Accordingly, awareness as well as acknowledgement of fear of emotions and bodily symptoms experienced by AYA cancer survivors may be of importance when providing psychological interventions to the population.

Imbalance in activity

The third clinical implication of study findings relates to AYAs cancer survivors reporting imbalance in activity, which is consistent with the wider literature (Krull et al., 2010). Typical concerns reported were identity difficulties, fear of failure, and lack of routines, with maintaining factors such as lack of engagement with positively reinforcing activities, maladaptive stress behaviors, and procrastination. Activity restriction is associated with impaired mental (Leventhal, 2008) and physical health (Kodama et al., 2009) and may increase the prevalence of late effects in the population (Lipshultz et al., 2015). Whilst low levels of physical activity in AYA cancer survivors are reported, (Bélanger et al., 2011; Hocking et al., 2013) less is known regarding AYA cancer survivors' disengagement from positively reinforcing activities in other areas. Focusing on increasing opportunities for engagement in positively reinforcing activities, an evidence-based psychological approach like behavioral activation (Ekers et al., 2014) may represent a solution for AYA cancer survivors. Furthermore, behavioral activation can be combined with physical activity promotion (Euteneuer et al., 2017; Pentecost et al., 2015) thus potentially targeting both mental and physical health.

Worry and rumination

In line with a pilot trial of Meta Cognitive Therapy for AYA cancer survivors (Fisher et al., 2015), our findings identified worry and rumination as potentially important targets in the provision of psychological interventions for the population. Key characteristics were worry about

the future and fear of cancer recurrence, causing decisional anxiety and depressive as well as existential rumination. Indeed, other research has shown AYA cancer survivors to imagine their future in more illness focused and overgeneralized way, in comparison to AYAs without cancer (Sansom-Daly et al., 2018). Given adaptive problem solving and goal setting are associated with the ability to imagine future events more specifically (Schacter, Addis & Buckner, 2007) such overgeneralized thinking may be an important target for psychological interventions for the AYA cancer survivor population (Sansom-Daly et al., 2018).

Limitations

First, due to most of those approached refraining from study participation, a resultant small sample hampered possibilities of examining potential differences between AYA cancer survivors who were recruited into the study, versus those who declined participation. Still, a preliminary evaluation was conducted and findings highlight important feasibility concerns in the recruitment of AYA cancer survivors. In addition, a number of important feasibility outcomes were not examined, for example, intervention acceptability, acceptability of study outcome measurements, safety, study resources, cost, and therapist training needs and competence (Eldridge et al., 2016; Sansom-Daly et al., 2018). Future studies may benefit from examining a wider range of feasibility outcomes. Further, an a priori set of progression criteria to assess trial feasibility was not established. This is a common limitation in feasibility and pilot trials (Mbuagbaw et al., 2019), resulting in it being difficult to interpret the findings of the present study. When interpreting the cognitive behavioral conceptualization, the impact of study therapists' and researchers' background, clinical experience, and theoretical orientation on data collection and analysis should be considered. No formal assessment of therapist competence to the validity of the behavioral case formulations was conducted, and individual case formulations were not empirically evaluated (Mumma & Fluck, 2016). Further, therapist adherence to the CBT model was not assessed. This is of particular importance given fidelity to the intervention may be associated with study outcomes and assist interpretation of findings (Mars et al., 2013). As such, the presented conceptualization of cancer-related concerns and maintaining factors is a hypothesis that could be examined in future studies, whom may benefit from exploring alternative approaches to conceptualizing distress within the population. In addition, member checking (Lincoln & Guba, 1985) to ensure themes generated and subsequent interpretations resonated with interviewees' experiences was not conducted but could have increased trustworthiness (Krefting, 1991). Further checking to establish trustworthiness with oncology health professionals and parents of AYA cancer survivors could have been conducted. However, it should be noted that a variety of techniques were utilized to establish trustworthiness, including triangulation of both data sources and investigators (Knafl & Breitmayer, 1989). It is unclear whether theoretical saturation and redundancy of data were met (Johnson, Hayes & Wade, 2007). However, given the diversity of human experience, theoretical saturation and redundancy of data may never be achieved and trustworthiness cannot be determined by sample size (Williams & Morrow, 2009). Further research should be undertaken to examine the efficacy

of psychological treatment in targeting the cancer-related concerns identified. Finally, although one of the strengths of the current study consists of its efforts to identify cancer-related concerns, it cannot be known whether the same difficulties would have arisen regardless of the cancer experience. Despite these limitations, we believe our findings offer valuable insight for a theoretical understanding of distress among AYA cancer survivors, and the development of psychological support specifically tailored towards this distress.

Future Clinical Directions

Identification and conceptualization of cancer-related concerns and potential maintaining factors may inform future clinical directions concerning the provision of psychological support to an AYA cancer survivor population. First, evidence increasingly suggests cultural adaptation of CBT is required to work with people from diverse backgrounds (Naeem, 2019). The definition of a cultural group, or subgroup, is not limited to race or religion, but includes factors such as age and physical health status (Naeem, 2019) and thus the AYA cancer survivor population may be considered a cultural group. Given different cultural groups may hold different beliefs concerning the cause and treatment of mental health difficulties and help seeking, it is important to appreciate specific concerns of an AYA cancer survivor population to ensure appropriate CBT adaptation. Indeed, the importance of patient-adapted CBT for the specific needs of an adult cancer population has been identified elsewhere (Poort et al., 2018).

Second, evidence based CBT models for the main difficulties identified in AYA cancer survivors exist. For example for social anxiety disorder (Clark & Wells, 1995; Rapee & Heimberg, 1997), health anxiety (Salkovskis & Warwick, 1986), and worry (Wells, 1999) and as such can inform the development of future interventions for the population. Evidence based CBT techniques for social anxiety may include, but are not limited to, behavioral experiments, attention training, and experiential exercises to understand the negative effects of safety behaviors (Leigh & Clark, 2018) and the addition of social skills training has been associated with improved therapeutic outcomes in children and adolescents (Scaini et al., 2016). Behavioral experiments and graded exposure (e.g., to avoided illness related situations) and response prevention are evidence based CBT techniques that may be utilized for health anxiety (Warwick et al., 1996). Further, behavioral activation is an evidence based technique (Ekers, 2014) for depression to facilitate re-engagement in positively reinforcing activities.

Third, findings suggest AYA cancer survivors experience a diverse range of difficulties and concerns, for example social anxiety, fear of cancer reoccurrence, worry and depression. Such findings are in line with high comorbidity rates between anxiety and depressive disorders in adolescent and young adult populations (Essau, 2008) and suggest interventions targeting clusters of symptoms, as opposed to individual diagnoses, (Ranøyen et al., 2018) may be more appropriate. A transdiagnostic conceptualization of distress and intervention approach (Gros, Allan & Szafranski, 2016), targeting underlying maintaining processes (e.g., experiential avoidance and rumination) as opposed to diagnosis specific symptoms (Norton & Paulus, 2016),

may be more appropriate (Ander et al., 2018). Indeed, transdiagnostic CBT interventions have shown promise for an adolescent population (Ehrenreich-May et al., 2017) and can be administered via the internet (Newby et al., 2015). Future research may wish to examine the promise of transdiagnostic CBT for an AYA cancer survivor population, including internet-administered interventions.

Conclusions

Whilst able to reveal a number of cancer-related concerns and a conceptualization of these for AYA cancer survivors, our study suffered from a poor recruitment rate, suggesting a need to understand help-seeking behavior in the population. Though there was indication of clinical effectiveness of the intervention used, caution should be taken due to the small sample size. Alternative recruitment strategies for the population appear called for, such as using social media and other internet-based methods. Given practical barriers seen in the population such as lack of time and difficulties of scheduling and attending intervention sessions, internet-administered alternatives may also be more appropriate in terms of support and intervention delivery, in offering larger flexibility than individualized face-to-face CBT. Finally, present results of cancer-related concerns experienced by AYA cancer survivors may prove a step in informing the development of psychological support, tailored to their unique needs.

Acknowledgements

We are grateful to Lisa Ljungman for her work as therapists. We would also like to thank Annika Lindahl Norberg for the supervision of qualitative interviews, as well as therapist supervisors Cecilia Arlinger Karlsson and Peter Czatlos.

References

Abrams AN, Hazen EP, Penson RT. 2007. Psychosocial issues in adolescents with cancer. *Cancer Treatment Reviews* 33(7):622-630. DOI: 10.1016/j.ctrv.2006.12.006.

American Psychiatric Association. 2000. *Diagnostic and Statistical Manual of Mental Disorders.* 4th ed. Washington, DC: American Psychiatric Association.

Ander M, Grönqvist H, Cernvall M, Engvall G, Hedström M, Ljungman G, Lyhagen J, Mattsson E, von Essen L. 2016. Development of health-related quality of life and symptoms of anxiety and depression among persons diagnosed with cancer during adolescence: a 10-year follow-up study. *Psycho-Oncology* 25(5):582-589. DOI: 10.1002/pon.3965.

Ander M, Thorsell Cederberg J, von Essen L, Hovén E. 2018. Exploration of psychological distress experienced by survivors of adolescent cancer reporting a need for psychological support. *PLoS ONE* 13(4): e0195899. DOI: 10.1371/journal.pone.0195899.

Barnett M, McDonnell G, DeRosa A, Schuler T, Philip E, Peterson L, Touza K, Jhanwar S, Atkinson TM, Ford JS. 2016. Psychosocial outcomes and interventions among cancer survivors diagnosed during adolescence and young adulthood (AYA): a systematic review. *Journal of Cancer Survivorship* 10(5):814-31. DOI: 10.1007/s11764-016-0527-6.

Beck AT, Epstein N, Brown G, Steer RA. 1988. An inventory for measuring clinical anxiety: psychometric properties. *Journal of Consulting and Clinical Psychology* 56(6):893-897. DOI: 10.1037/0022-006X.56.6.893.

Bélanger LJ, Plotnikoff RC, Clark A, Courneya KS. 2011. Physical activity and health-related quality of life in young adult cancer survivors: a Canadian provincial survey. *Journal of Cancer Survivorship* 5(1):44-53. DOI: 10.1007/s11764-010-0146-6.

Bibby H, White V, Thompson K, Anazodo A. 2017. What are the unmet needs and care experiences of adolescents and young adults with cancer? A systematic review. *Journal of Adolescent and Young Adult Oncology* 6(1):6-30. DOI: 10.1089/jayao.2016.0012.

Billingham SAM, Whitehead AL, Julious SA. 2013. An audit of sample sizes for pilot and feasibility trials being undertaken in the United Kingdom registered in the United Kingdom Clinical Research Network database. *BMC Medical Research Methodology* 13:104. DOI: 10.1186/1471-2288-13-104.

Bond FW, Hayes SC, Baer RA, Carpenter KM, Guenole N, Orcutt HK, Waltz T, Zettle RD. 2011. Preliminary psychometric properties of the Acceptance and Action Questionnaire-II: a revised measure of psychological inflexibility and experiential avoidance. *Behavior Therapy* 42(4):676-688. DOI: 10.1016/j.beth.2011.03.007.

Braun V, Clarke V. 2006. Using thematic analysis in psychology. *Qualitative Research in Psychology* 3(2):77-101.

Breuer N, Sender A, Daneck L, Mentschke L, Leuteritz K, Friedrich M, Nowe E, Stöbel-Richter Y, Geue K. 2017. How do young adults with cancer perceive social support? A qualitative study. *Journal of Psychosocial Oncology* 35(3):292-308. DOI: 10.1080/07347332.2017.1289290.

Campo RA, Bluth K, Santacroce SJ, Knapik S, Tan J, Gold S, Philips K, Gaylord S, Asher GN. 2017. A mindful self-compassion videoconference intervention for nationally recruited posttreatment young adult cancer survivors: feasibility, acceptability, and psychosocial outcomes. *Supportive Care in Cancer* 25(6):1759-1768. DOI: 10.1007/s00520-017-3586-y.

Cho D, Park CL. 2017. Moderating effects of perceived growth on the association between fear of cancer recurrence and health-related quality of life among adolescent and young adult cancer survivors. *Journal of Psychosocial Oncology* 35(2):148-165. DOI: 10.1080/07347332.2016.1247408.

Clark DM, Wells A. 1995. A cognitive model of social phobia. In: Heimberg RG, Liebowitz MR, Hope DA, Schneier FR, eds. *Social Phobia: Diagnosis, Assessment, and Treatment*. New York, NY: Guilford Press, pp 69-93.

Clement S, Schauman O, Graham T, Maggioni F, Evans-Lacko S, Bezborodovs N, Morgan C, Rüsch N, Brown JS, Thornicroft G. 2015. What is the impact of mental health-related stigma on help-seeking? A systematic review of quantitative and qualitative studies. *Psychological Medicine* 45(1):11-27. DOI: 10.1017/S0033291714000129

Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M. 2008. Developing and evaluating complex interventions: the new Medical Research Council guidance. *British Medical Journal* 337:a1655. DOI: 10.1136/bmj.a1655.

De Laage A, Allodji R, Dauchy S, Rivollet S, Fayech C, Fresneau B, Oberlin O. 2016. Screening for psychological distress in very long-term adult survivors of childhood cancer. *Pediatric Hematology and Oncology* 33(5):295-313. DOI: 10.1080/08880018.2016.1204400

Devine KA, Viola AS, Coups EJ, Wu YP. 2018. Digital health interventions for adolescent and young adult cancer survivors. *JCO Clinical Cancer Informatics* 2:1-15. DOI: 10.1200/CCI.17.00138.

Dougher MJ, ed. 2000. *Clinical Behaviour Analysis*. Reno: Context Press.

Ehrenreich-May J, Rosenfield D, Queen AH, Kennedy SM, Remmes CS, Barlow DH. 2017. An initial waitlist-controlled trial of the unified protocol for the treatment of emotional disorders in adolescents. *Journal of Anxiety Disorders* 46:46-55. DOI: 10.1016/j.janxdis.2016.10.006.

Ekers D, Webster L, Van Straten A, Cuijpers P, Richards D, Gilbody S. 2014. Behavioural activation for sepression: an update of meta-analysis of effectiveness and sub group analysis. *PLOS One* 9(6):e100100. DOI: 10.1371/journal.pone.0100100.

Eldridge SM, Lancaster GA, Campbell MJ, Thabane L, Hopewell S, Coleman CL, Bond CM. 2016. Defining feasibility and pilot studies in preparation for randomised controlled trials: development of a conceptual framework. *PLoS One* 11(3):e0150205. DOI: 10.1371/journal.pone.0150205.

Essau CA. 2008. Comorbidity of depressive disorders among adolescents in community and clinical settings. *Psychiatry Research* 158(1), 35-42. DOI: 10.1016/j.psychres.2007.09.007.

Euteneuer F, Dannehl K, del Rey A, Engler H, Schedlowski M, Rief1 W. 2017. Immunological effects of behavioral activation with exercise in major depression: an exploratory randomized controlled trial. *Translational Psychiatry* 7(5): e1132. DOI: 10.1038/tp.2017.76.

Feilzer MY. 2009. Doing mixed methods research pragmatically: implications for the rediscovery of pragmatism as a research paradigm. *Journal of Mixed Methods Research* 4(1):6-16. DOI: 10.1177/1558689809349691

Fisher PL, McNicol K, Young B, Smith E, Salmon P. 2015. Alleviating emotional distress in adolescent and young adult cancer survivors: an open trial of metacognitive therapy. *Journal of Adolescent and Young Adult Oncology* 4(2):64-69. DOI: 10.1089/jayao.2014.0046.

Fydrich T, Dowdall D, Chambless DL. 1992. Reliability and validity of the Beck Anxiety Inventory. *Journal of Anxiety Disorders* 6(1):55-61. DOI: 10.1016/0887-6185(92)90026-4.

Gorman JR, Roberts SC, Dominick SA, Malcarne VL, Dietz AC, Su HI. 2014. A diversified recruitment approach incorporating social media leads to research participation among young adult-aged female cancer survivors. *Journal of Adolescent and Young Adult Oncology* 3(2):59-65. DOI: 10.1089/jayao.2013.003.

737 Gregory AM, Caspi A, Moffitt TE, Koenen K, Eley TC, Poulton R. 2007. Juvenile mental health
738 histories of adults with anxiety disorders. *American Journal of Psychiatry* 164(2):301-8.
739 DOI: 10.1176/ajp.2007.164.2.301.
740 Gros DF, Allan NP, Szafranski DD. 2016. Movement towards transdiagnostic psychotherapeutic
741 practices for the affective disorders. *Evidence-Based Mental Health* 19(3):e10-2. DOI:
742 10.1136/eb-2015-102286.
743 Gulliver A, Griffiths KM, Christensen H. 2010. Perceived barriers and facilitators to mental
744 health help-seeking in young people: a systematic review. *BMC Psychiatry* 10:113. DOI:
745 10.1186/1471-244X-10-113.
746 Gunnes MW, Lie RT, Bjørge T, Ghaderi S, Syse A, Ruud E, Wesenberg F, Moster D. 2017.
747 Suicide and violent deaths in survivors of cancer in childhood, adolescence and young
748 adulthood - A national cohort study. *International Journal of Cancer* 140(3):575-580.
749 DOI: 10.1002/ijc.30474.
750 Haase JE. 2004. The Adolescent Resilience Model as a guide to interventions. *Journal of*
751 *Pediatric Oncology Nursing* 21(5):289-299. DOI: 10.1177/1043454204267922.
752 Hayes SC, Wilson K., Gifford EV, Follette VM, Strosahl K. 1996. Experiental avoidance and
753 behavioral disorders: A functional dimensional approach to diagnosis and treatment.
754 *Journal of Consulting Clinical Psychology* 64(6):1152-1168. DOI: 10.1037/0022-
755 006X.64.6.1152
756 Hocking MC, Schwartz LA, Hobbie WL, Derosa BW, Ittenbach RF, Mao JJ, Ginsberg JP,
757 Kazak AE. 2013. Prospectively examining physical activity in young adult survivors of
758 childhood cancer and healthy controls. *Pediatric Blood and Cancer* 60(2):309-15. DOI:
759 10.1002/pbc.24144.
760 Hollis C, Morriss R, Martin J, Amani S, Cotton R, Denis M, Lewis M. 2015. Technological
761 innovations in mental healthcare: harnessing the digital revolution. *British Journal of*
762 *Psychiatry* 206(4):263-5. DOI: 10.1192/bjp.bp.113.142612.
763 Hopwood P, Fletcher I, Lee A, Al Ghazal S. 2001. A body image scale for use with cancer
764 patients. *European Journal of Cancer* 37:189-197. DOI: 10.1016/S0959-8049(00)00353-1.
765 Husson O, Zebrack BJ, Aguilar C, Hayes-Lattin B, Cole S. 2017. Cancer in adolescents and
766 young adults: Who remains at risk of poor social functioning over time? *Cancer*
767 123(14):2743-2751. DOI: 10.1002/cncr.30656.
768 Jacobson NS, Truax P. 1991. Clinical significance: a statistical approach to defining meaningful
769 change in psychotherapy research. *Journal of Consulting and Clinical Psychology*
770 59(1):12-19. DOI: 10.1037/0022-006X.59.1.12.
771 Jaworska N, MacQueen G. 2015. Adolescence as a unique developmental period. *Journal of*
772 *Psychiatry and Neuroscience* 40(5):291-293. DOI: 10.1503/jpn.150268.
773 Johannsdottir IM, Loge JH, Kiserud CE, Karlstad Ø, Skurtveit S. 2018. Increased prescription
774 rates of anxiolytics and hypnotics to survivors of cancer in childhood, adolescence, and
775 young adulthood-A population-based study. *Pediatric Blood and Cancer* 65(2). DOI:
776 10.1002/pbc.26860.

777 Johnson CV, Hayes JA, Wade NG. 2007. Psychotherapy with troubled spirits: A qualitative
778 investigation. *Psychotherapy Research* 17(4):450-460. DOI:
779 10.1080/10503300600953520.
780 Kaczkurkin AN, Foa EB. 2015. Cognitive-behavioral therapy for anxiety disorders: an update on
781 the empirical evidence. *Dialogues in Clinical Neuroscience* 17(3):337-346.
782 Kaul S, Avila JC, Mutambudzi M, Russell H, Kirchhoff AC, Schwartz CL. 2017. Mental distress
783 and health care use among survivors of adolescent and young adult cancer: A cross-
784 sectional analysis of the National Health Interview Survey. *Cancer* 123(5):869-878. DOI:
785 10.1002/cncr.30417.
786 Kazak AE, Alderfer M, Rourke MT, Simms S, Streisand R, Grossman JR. 2004. Posttraumatic
787 Stress Disorder (PTSD) and Posttraumatic Stress Symptoms (PTSS) in families of
788 adolescent childhood cancer survivors. *Journal of Pediatric Psychology* 29(3):211-219.
789 DOI: 10.1093/jpepsy/jsh022.
790 Kazak AE, Derosa BW, Schwartz LA, Hobbie W, Carlson C, Ittenback RF, Mao JJ, Ginsberg JP.
791 2010. Psychological outcomes and health beliefs in adolescent and young adult survivors
792 of childhood cancer and controls. *Journal of Clinical Oncology* 28(12):2002-2007. DOI:
793 10.1200/JCO.2009.25.9564.
794 Kazak AE, Kassam-Adams N, Schneider S, Zelikovsky N, Alderfer MA, Rourke M. 2006. An
795 integrative model of pediatric medical traumatic stress, *Journal of Pediatric Psychology*,
796 31(4):343-355. DOI: 10.1093/jpepsy/jsj054.
797 Kessler RC, Avenevoli S, Costello J, Green JG, Gruber MJ, McLaughlin KA, Petukhova M,
798 Sampson NA, Zaslavsky AM, Merikangas KR. 2012. Severity of 12-month DSM-IV
799 disorders in the National Comorbidity Survey Replication Adolescent Supplement.
800 *Archives of General Psychiatry* 69(4):381-389. DOI:
801 10.1001/archgenpsychiatry.2011.1603.
802 Kilsdonk E, van Dulmen-den Broeder E, van der Pal HJ, Hollema N, Kremer LC, van den
803 Heuvel-Eibrink MM, van Leeuwen FE, Jaspers MW, van den Berg MH. 2015. Effect of
804 web-based versus paper-based questionnaires and follow-up strategies on participation
805 rates of Dutch childhood cancer survivors: a randomized controlled trial. *Journal of*
806 *Medical Internet Research Cancer* 1(2):e11. DOI: 10.2196/cancer.3905.
807 Knafl KA, Breitmayer, BJ. 1989. *Triangulation in qualitative research: Issues of conceptual*
808 *clarity and purpose.* In J. Morse (Ed.), Qualitative nursing research: A contemporary
809 dialogue (pp. 193-203). Rockville, MD: Aspen.
810 Kodama S, Saito K, Tanaka S, Maki M, Yachi Y, Asumi M, Sugawara A, Totsuka K, Shimano
811 H, Ohashi Y, Yamada N, Sone H. 2009. Cardiorespiratory fitness as a quantitative
812 predictor of all-cause mortality and cardiovascular events in healthy men and women.
813 *JAMA* 301(19):2024-2035. DOI: 10.1001/jama.2009.681.
814 Kovandžić M, Chew-Graham C, Edwards S, Peters S, Edge D, Aseem S, Gask L, Dowrick C.
815 2011. Access to primary mental health care for hard-to-reach groups: from 'silent suffering'

to 'making it work'. *Social Science & Medicine* 72(5):763-772. DOI: 10.1016/j.socscimed.2010.11.027.

Krefting L. 1991. Rigor in qualitative research: the assessment of trustworthiness. *American Journal of Occupational Therapy* 45(3):214-22. DOI:10.5014/ajot.45.3.214

Kroenke K, Spitzer RL, Williams JBW. 2002. The PHQ-15: validity of a new measure for evaluating the severity of somatic symptoms. *Psychosomatic Medicine* 64(2):258-266. DOI10.1097/00006842-200203000-00008.

Krull KR, Huang S, Gurney JG, Klosky JL, Leisenring W, Termuhlen A, Ness KK, Srivastava DK, Mertens A, Stovall M, Robison LL, Hudson MM. 2010. Adolescent behavior and adult health status in childhood cancer survivors. *Journal of Cancer Survivorship* 4(3):210-217. DOI: 10.1007/s11764-010-0123-0.

Lang MJ, Giese-Davis, J, Patton SB, Campbell, DJT. 2018. Does age matter? Comparing post-treatment psychosocial outcomes in young adult and older adult cancer survivors with their cancer-free peers. *Psycho-Oncology* 27(5):1404–1411. DOI: 10.1002/pon.4490.

Larsson G, Mattsson E, von Essen L. 2010. Aspects of quality of life, anxiety, and depression among persons diagnosed with cancer during adolescence: a long-term follow-up study. *European Journal of Cancer* 46(6):1062-1068. DOI: 10.1016/j.ejca.2010.01.021.

Lehmann V, Gronqvist H, Engvall G, Ander M, Tuinman MA, Hagedoorn M, Sanderman R, Mattsson E, von Essen L. 2014. Negative and positive consequences of adolescent cancer 10 years after diagnosis: an interview-based longitudinal study in Sweden. *Psycho-Oncology* 23(11):1229-1235. DOI: 10.1002/pon.3549.

Leigh E, Clark DM. 2018. Understanding social anxiety disorder in adolescents and improving treatment outcomes: applying the Cognitive Model of Clark and Wells (1995). *Clin Child Fam Psychol Rev* 21(3):388-414. DOI: 10.1007/s10567-018-0258-5.

Leventhal AM. 2008. Sadness, depression, and avoidance behavior. *Behavior Modification* 32(6):759-779. DOI: 10.1177/0145445508317167.

Lincoln YS, Guba EA. 1985. *Naturalistic Inquiry*. Beverly Hills, CA: Sage.

Lipshultz SE, Franco VI, Miller TL, Colan SD, Sallan SE. 2015. Cardiovascular disease in adult survivors of childhood cancer. *Annual Review of Medicine* 66:161-176. DOI:10.1146/annurev-med-070213-054.

Mars T, Ellard D, Carnes D, Horner K, Underwoord M, Taylor SJC. 2013. Fidelity in complex behaviour change interventions: a standardised approach to evaluate intervention integrity. *BMJ Open* 3:e003555. DOI: 10.1136/bmjopen-2013-003555.

Mbuagbaw L, Kosa SD, Lawson DO, Stalteri R, Olaiya OR, Alotaibi A, Thabane L. 2019. The reporting of progression criteria in protocols of pilot trials designed to assess the feasibility of main trials is insufficient: a meta-epidemiological study. *Pilot and Feasibility Studies* 5:120. DOI: 10.1186/s40814-019-0500-z.

McCusker J, Yaffe M, Faria R, Lambert S, Li M, Poirier-Bisson J, Magalhaes M, de Raad M. 2018. Phase II trial of a depression self-care intervention for adult cancer survivors. *European Journal of Cancer Care* 27(1):e12763. DOI: 10.1111/ecc.12763.

McLaughlin KA & Hatzenbuehler ML. 2009. Stressful life events, anxiety sensitivity, and internalizing symptoms in adolescents. *Journal of Abnormal Psychology* 118(3):659-669. DOI: 10.1037/a0016499.

McMain S, Newman MG, Segal ZV, DeRubeis RJ. 2015. Cognitive behavioral therapy: current status and future research directions. *Psychotherapy Research* 25(3):321-329. DOI: 10.1080/10503307.2014.1002440.

Mesa F, Le TA, Beidel DC. 2015. Social skill-based treatment for social anxiety disorder in adolescents. In: Ranta K, La Greca A, Garcia-Lopez LJ, Marttunen M, ed. *Social Anxiety and Phobia in Adolescents*. Springer, Cham, 289-299.

Meyer TJ, Miller ML, Metzger RL, Borkovec TD. 1990. Development and validation of the Penn State Worry Questionnaire. *Behaviour Research and Therapy* 28(6):487-495. DOI: 10.1016/0005-7967(90)90135-6.

Michielsen HJ, de Vries J, van Heck GL. 2003. Psychometric qualities of a brief self-rated fatigue measure: the Fatigue Assessment Scale. *Journal of Psychosomatic Research* 54(4):345-352. DOI: 10.1016/S0022-3999(02)00392-6.

Moher D, Hopewell S, Schulz KF, Montori V, Gøtzsche PC, Devereaux PJ, Elbourne D, Egger M, Altman DG. 2010. CONSORT 2010 explanation and elaboration: updated guidelines for reporting parallel group randomised trials. *British Medical Journal* 340:c869. DOI: 10.1136/bmj.c869.

Moody L, Turner A, Osmond J, Hooker L, Kosmala-Anderson J, Batehup L. 2015. Web-based self-management for young cancer survivors: consideration of user requirements and barriers to implementation. *Journal of Cancer Survivorship* 9(2):188–200. DOI: 10.1007/s11764-014-0400-4.

Mumma G, Fluck J. 2016. How valid is your case formulation? Empirically testing your cognitive behavioural case formulation for tailored treatment. *The Cognitive Behavioral Therapist* 9:1-25. DOI: 10.1017/S1754470X16000088

Musiat P, Winsall M, Orlowski S, Antezana G, Schrader G, Battersby M, Bidargaddi N. 2016. Paid and Unpaid Online Recruitment for Health Interventions in Young Adults. *Journal of Adolescent Health*, 59(6):662-667. DOI: 10.1016/j.jadohealth.2016.07.020.

Naeem, F. 2019. Cultural adaptations of CBT: a summary and discussion of the Special Issue on Cultural Adaptation of CBT. *The Cognitive Behaviour Therapist*, 12, e40. DOI: 10.1017/S1754470X19000278P.

Newby JM, McKinnon A, Kuyken W, Gilbody S, Daigleish T. 2015. Systematic review and meta-analysis of transdiagnostic psychological treatments for anxiety and depressive disorders in adulthood. *Clinical Psychology Review* 40:91-110. DOI: 10.1016/j.cpr.2015.06.002.

Nolen-Hoeksema S. 1991. Responses to depression and their effects on the duration of depressive episodes. *Journal of Abnormal Psychology* 100(4):569-582. DOI:10.1037/0021-843X.100.4.569.

Norton P, Paulus D. 2016. Toward a unified treatment for emotional disorders: update on the science and practice. *Behavior Therapy* 47(6):854-868. DOI: 10.1016/j.beth.2015.07.002.

Pentecost C, Farrand P, Greaves CJ, Taylor RS, Warren FC, Hillsdon M, Green C, Welsman JR, Rayson K, Evans PH, Taylor AH. 2015. Combining behavioural activation with physical activity promotion for adults with depression: findings of a parallel-group pilot randomised controlled trial (BAcPAc). *Trials* 16:367. DOI: 10.1186/s13063-015-0881-0.

Persons JB. 2008. *The case formulation approach to cognitive-behaviour therapy*. New York, NY: Guilford Press.

Poort H, Meade CD, Knoop H, Gielissen MFN, Pinilla-Ibarz J, Jacobsen PB. 2018. Adapting an evidence-based intervention to address targeted therapy-related fatigue in chronic myeloid leukemia patients. *Cancer Nursing*, 41(1), e28-e37. DOI: 10.1097/NCC.0000000000000446.

Rabin C, Horowitz S, Marcus B. 2013. Recruiting young adult cancer survivors for behavioral research. *Journal of Clinical Psychology in Medical Settings* 20(1):33-36. DOI: 10.1007/s10880-012-9317-0.

Ranøyen I, Lydersen S, Larose TL, Weidle B, Skokauskas N, Thomsen PH, Wallander J, Indredavik MS. 2018. Developmental course of anxiety and depression from adolescence to young adulthood in a prospective Norwegian clinical cohort. *European Child & Adolescent Psychiatry* 27(11):1143-1423. DOI: 10.1007/s00787-018-1139-7.

Rapee RM, Heimberg RG. 1997. A cognitive-behavioral model of anxiety in social phobia. *Behaviour Research and Therapy* 35:741-56. DOI: 10.1016/s0005-7967(97)00022-3

Richards DA, Hill JJ, Gask L, Lovell K, Chew-Graham C, Bower P, Cape J, Pilling S, Araya R, Kessler D, Bland JM, Green C, Gilbody S, Lewis G, Manning C, Hughes-Morley A, Barkham M. 2013. Clinical effectiveness of collaborative care for depression in UK primary care (CADET): cluster randomised controlled trial. *BMJ Open* 347:f4913. DOI: 10.1136/bmj.f4913.

Richards DA, Ekers D, McMillan D, Taylor RS, Byford S, Warren FC, Barrett B, Farrand PA, Gilbody S, Kuyken W, O'Mahen H, Watkins ER, Wright KA, Hollon SD, Reed N, Rhodes S, Fletcher E, Finning K. 2016. Cost and Outcome of Behavioural Activation versus Cognitive Behavioural Therapy for Depression (COBRA): a randomised, controlled, non-inferiority trial. *Lancet* 388(10047):871-80. DOI: 10.1016/S0140-6736(16)31140-0.

Rickwood D, Deane FP, Wilson CJ Ciarrochi JV. 2005. Young people's help-seeking for mental health problems. *Australian e-Journal for the Advancement of Mental Health*, 4(3):1-34.

Ritchie J, Spencer L. 1994. Qualitative Data Analysis for Applied Policy Research. In: Bryman, A. and Burgess, R., Eds., *Analyzing Qualitative Data*, Routledge, London, 173-194.

Robb SL, Burns DS, Stegenga KA, Haut PR, Monahan PO, Meza J, Stump TE, Cherven BO, Docherty SL, Hendricks-Ferguson VL, Kintner EK, Haight AE, Wall DA, Haase JE. 2014. Randomized clinical trial of therapeutic music video intervention for resilience outcomes in adolescents/young adults undergoing hematopoietic stem cell transplant: A report from the Children's Oncology Group. *Cancer* 120(6):909-917. DOI: 10.1002/cncr.28355

Roth ME, O'Mara AM, Seibel NL, Dickens DS, Langevin AM, Pollock BH, Freyer DR. 2016. Low enrollment of adolescents and young adults onto cancer trials: Insights from the Community Clinical Oncology Program. *Journal of Oncology Practice* 12(4):e388-e395. DOI: 10.1200/JOP.2015.009084.

Ruggiero KJ, Ben KD, Scotti JR, Rabalais AE. 2003. Psychometric properties of the PTSD checklist—civilian version. *Journal of Traumatic Stress* 16(5):495-502. DOI: 10.1023/A:1025714729117.

Salaheddin K, Mason B. 2016. Identifying barriers to mental health help-seeking among young adults in the UK: a cross-sectional survey. *British Journal of General Practice* 66:e686-92. DOI: 10.3399/bjgp16X687313.

Salkovskis PM, Rimes KA, Warwick HMC, Clark DM. 2002. The Health Anxiety Inventory: development and validation of scales for the measurement of health anxiety and hypochondriasis. *Psychological Medicine* 32(5):843-853. DOI: 10.1017\S0033291702005822.

Salkovskis PM, Warwick HMC. 1986. Morbid preoccupations, health anxiety and reassurance: A cognitive-behavioural approach to hypochondriasis. *Behav Res Ther* 24:597–602. DOI: 10.1016/0005-7967(86)90041-0

Sansom-Daly UM, Evans HE, Ellis SJ, McGill BC, Hetherington K, Wakefield CE. 2017. Something's got to give: time–cost trade-offs in site-specific research approval can negatively impact patient recruitment in multi-institutional studies. *Internal Medicine Journal* 47:1088-1089. DOI: 10.1111/imj.13540.

Sansom-Daly UM, Wakefield CE, Bryant RA, Patterson P, Anazodo A, Butow P, Sawyer S, McGill BC, Evans H, Cohn RJ, and The Recapture Life Working Party. 2018. Feasibility, acceptability, and safety of the Recapture Life videoconferencing intervention for adolescent and young adult cancer survivors. *Psycho-Oncology* 28:284-292. DOI: 10.1002/pon.4938.

Sansom-Daly UM, Wakefield CE. 2013. Distress and adjustment among adolescents and young adults with cancer: An empirical and conceptual review. *Translational Pediatrics*, 2(4):167-197. DOI: 10.3978/j.issn.2224-4336.2013.10.06.

Scaini S, Belotti R, Ogliari A, Battaglia M. 2016. A comprehensive meta-analysis of cognitive-behavioral interventions for social anxiety disorder in children and adolescents. *Journal of Anxiety Disorders* 42:105-12. DOI: 10.1016/j.janxdis.2016.05.008.

Schacter DL, Addis DR, Buckner RL. 2007. Remembering the past to imagine the future: the prospective brain. *Nature reviews Neuroscience* 8(9):657-661. DOI: 10.1038/nrn2213.

Schulte F. 2015. Social competence in pediatric brain tumor survivors. *Current Opinion in Oncology* 27(4), 306–310. DOI:10.1097/cco.0000000000000193.

Seitz DC, Besier T, Debatin KM, Grabow D, Dieluweit U, Hinz A, Kaatsch P, Goldbeck L. 2010. Posttraumatic stress, depression and anxiety among adult long-term survivors of cancer in adolescence. *European Journal of Cancer* 46(9):1596-1606. DOI: 10.1016/j.ejca.2010.03.001.

Seitz DC, Besier T, Goldbeck L. 2009. Psychosocial interventions for adolescent cancer patients: a systematic review of the literature. *Psycho-Oncology* 18(7):683-690. DOI: 10.1002/pon.1473.

Seitz DC, Knaevelsrud C, Duran G, Waadt S, Loos S, Goldbeck L. 2014. Efficacy of an internet-based cognitive-behavioral intervention for long-term survivors of pediatric cancer: a pilot study. *Supportive Care in Cancer* 22(8):2075-2083. DOI: 10.1007/s00520-014-2193-4.

Sheehan DV, Lecrubier Y, Sheehan KH, Amorim P, Janavs J, Weiller E, Hergueta T, Baker R, Dunbar GC. 1998. The Mini-International Neuropsychiatric Interview (M.I.N.I.): the development and validation of a structured diagnostic psychiatric interview for DSM-IV and ICD-10. *Journal of Clinical Psychiatry* 59(Suppl 20):22-33.

Sheehan DV. 1983. *The Anxiety Disease.* New York: Charles Scribner & Sons.

Spinhoven P, van Hemert AM, Penninx BWJH. 2017. Experiential avoidance and bordering psychological constructs as predictors of the onset, Relapse and Maintenance of Anxiety Disorders: One or Many? *Cognitive Therapy Research* 41:867-880. DOI: 10.1007/s10608-017-9856-7.

Steineck A, Bradford MC, Lau N, Scott S, Yi-Frazier JP, Rosenberg AR. 2019. A psychosocial intervention's impact on quality of life in AYAs with cancer: a post hoc analysis from the promoting resilience in stress management (PRISM) randomized controlled trial. *Children* 6(11):E124. DOI: 10.3390/children6110124.

Sturmey P. 2008. *Behavioural Case Formulation and Intervention: A Functional Analytic Approach.* Chichester, West Sussex: John Wiley & Sons.

Svanborg P, Åsberg M. 1994. A new self-rating scale for depression and anxiety states based on the Comprehensive Psychopathological Rating Scale. *Acta Psychiatrica Scandinavica* 89(1):21-28. DOI: 10.1111/j.1600-0447.1994.tb01480.x.

Tanielian T, Jaycox LH, Paddock SM, Chandra A, Meredith LS, Burnam MA. 2009. Improving treatment seeking among adolescents with depression: Understanding readiness for treatment. *Journal of Adolescent Health* 45(5):490-498. DOI: 10.1016/j.jadohealth.2009.03.005.

Thewes B, Kaal SEJ, Custers JAE, Manten-Horst E, Jansen R, Servaes P, van der Graaf WTA, Prins JB, Husson O. 2017. Prevalence and correlates of high fear of cancer recurrence in late adolescents and young adults consulting a specialist adolescent and young adult (AYA) cancer service. *Support Care Cancer* 26(5):1479-1487. DOI: 10.1007/s00520-017-3975-2.

Van der Gucht K, Takano K, Labarque V, Vandenabeele K, Nolf N, Kuylen S, Cosyns V, Van Broeck N, Kuppens P, Raes F. 2017. A mindfulness-based intervention for adolescents and young adults after cancer treatment: effects on quality of life, emotional distress, and cognitive vulnerability. *Journal of Adolescent and Young Adult Oncology* 6(2):307-317. DOI: 10.1089/jayao.2016.0070.

1012 Warner EL, Kent EE, Trevino KM, Parsons HM, Zebrack BJ, Kirchhoff AC. 2016. Social well-
1013 being among adolescents and young adults with cancer: a systematic review. *Cancer*
1014 122(7):1029-1037. DOI: 10.1002/cncr.29866.
1015 Warwick HM, Clark DM, Cobb AM, Salkovskis PM. 1996. A controlled trial of cognitive-
1016 behavioural treatment of hypochondriasis. *Br J Psychiatry* 169(2):189-95. DOI:
1017 10.1192/bjp.169.2.189
1018 Weathers FW, Litz BT, Herman DS, Huska JA, Keane TM. 1993. *The PTSD Checklist (PCL):*
1019 *reliability, validity, and diagnostic utility.* The Annual Meeting of International Society for
1020 Traumatic Stress Studies. San Antonio, TX.
1021 Wells A. 1999. A cognitive model of generalized anxiety disorder. *Behav Modif* 23(4):526-55.
1022 DOI: 10.1177/0145445599234002
1023 Wilford J, Buchbinder D, Fortier MA, Osann K, Shen V, Torno L, Sender LS, Parsons SK,
1024 Wenzel L. 2017. "She Was a Little Social Butterfly": a qualitative analysis of parent
1025 perception of social functioning in adolescent and young adult brain tumor survivors.
1026 *Journal of Pediatric Oncology Nursing* 34(4):239-249. DOI: 10.1177/1043454216688660.
1027 Williams EN, Morrow SL. 2009. Achieving trustworthiness in qualitative research: A pan-
1028 paradigmatic perspective. *Psychotherapy Research* 19(4-5):576-582. DOI:
1029 10.1080/10503300802702113.
1030 Younes N, Chollet A, Menard E, Melchior M. 2015. E-mental health care among young adults
1031 and help-seeking behaviors: a transversal study in a community sample. *Journal of*
1032 *Medical Internet Research* 17(5):e123. DOI: 10.2196/jmir.4254.
1033 Zimmermann P, Iwanski A. 2014. Emotion regulation from early adolescence to emerging
1034 adulthood and middle adulthood. *International Journal of Behavioral Development*
1035 38(2):182–194. DOI: 10.1177/0165025413515405.

Table 1(on next page)

Number of participants receiving the respective CBT-based treatment techniques in treatment.

Table 1:
Number of participants receiving the respective CBT-based treatment techniques in treatment.

Treatment technique	*n*
Functional analysis	10
Goal setting	10
Psychoeducation including experiential exercises	10
Setting a maintenance plan	9
Exploring values	8
Self-monitoring	8
Mindfulness-based exercises	7
Value-guided behavior change/exposure relating to social avoidance and fear of failure	6
Worry time, worry exposure, problem-solving	5
Behavioral activation	3
General affect exposure	3
Stress management techniques	3
Emotion regulation/distress tolerance training	2
Exposure (interoceptive exposure and exposure & response prevention)	2
Interpersonal effectiveness skills training	2
Relaxation exercises	1

Table 2(on next page)

Participant characteristics (*n*=10).

Note. N/*n* = number of participants; SD=Standard Deviation. [a]Self-reported late effects included anemia, balance problems, fatigue/tiredness, headaches, high blood pressure, kidney stone, loss of appetite, memory problems, nausea, nerve damage, nerve pain, numbness, speech difficulties, and thrombus.

Table 2:
Participant characteristics (*n*=10).

	Mean	SD	Range
Age at study-start (years)	21	2.9	17-25
Age at cancer diagnosis (years)	15.9	1.4	13-17
Time since end of cancer treatment (years) (*n*=9)	3.2	1.7	1-6
	n		
Female/Male	4/6		
Cancer diagnosis from self-report			
Leukemia	4		
CNS-tumor	2		
Lymphoma	2		
Soft tissue sarcoma	1		
Other malignancy	1		
Cancer diagnosis from registry			
Leukemia	5		
CNS-tumor	2		
Lymphoma	2		
Soft tissue sarcoma	1		
Other malignancy	0		
History of relapse yes/no	1/9		
Living situation			
With parents	6		
Not with parents	4		
Relationship status			
Single	5		
In a relationship	5		
Employment status			
Student	6		
Employed full-time	1		
Employed part-time	2		
Unemployed	1		
Sick leave	0		
Self-reported late effects yes[a]/no	8/2		
Previous psychological treatment yes/no	6/4		

Note. N/*n* = number of participants; SD=Standard Deviation.
[a]Self-reported late effects included anemia, balance problems, fatigue/tiredness, headaches, high blood pressure, kidney stone, loss of appetite, memory problems, nausea, nerve damage, nerve pain, numbness, speech difficulties, and thrombus

Table 3 (on next page)

Means, standard deviations, and ranges for the clinical outcome measurements at baseline, post-treatment, and three month follow-up assessment (*n*=10)

[a](*n*=8) *Note. n*=number of participants; SD=Standard Deviation; MADRS-S=Montgomery Åsberg Depression Rating Scale –Self Assessment; BAI=Beck Anxiety Inventory; PCL-C=The PTSD Checklist-Civilian Version; PSWQ=Penn State Worry Questionnaire; AAQ-II=Acceptance and Action Questionnaire-II; BIS=Body Image Scale; FAS=Fatigue Assessment Scale; PHQ-15=Public Health Questionnaire-15; R-RSQ=Rumination Scale of the Response Style Questionnaire; SDS=Sheehan Disability Scale; SHAI=The Short Health Anxiety Inventory.

Table 3:

Means, standard deviations, and ranges for the clinical outcome measurements at baseline, post-treatment, and three month follow-up assessment (*n*=10).

	Baseline (*n*=9)		Post (*n*=7)		Follow-up (*n*=8)	
	Mean	**SD (range)**	**Mean**	**SD (range)**	**Mean**	**SD (range)**
MADRS-S	17.0	6.2 (8-27)	10.9[a]	7.6 (3-26)	9.8	7.1 (3-25)
BAI	14.6	16.5 (1-56)	7.9[a]	6.5 (2-21)	6.1	8.0 (0-25)
PCL-C	37.0	14.2 (20-70)	27.1[a]	8.1 (20-45)	27.4	11.7 (18-55)
PSWQ	48.9	11.5 (32-63)	40.9[a]	5.7 (32-49)	38.8	9.3 (22.4-52)
AAQ-II	36.3	10.6 (23-55)	32.4[a]	10.3 (19-47)	28.6	12.1 (17-50)
BIS	13.2	7.9 (5-29)	8.0	4.1 (4-14)	8.8	8.1 (0-25)
FAS	26.8	5.6 (20-36)	22.3	5.5 (17-31)	21.5	5.6 (12-29)
PHQ-15	9.5	6.7 (2-23)	6.1	3.6 (1-12)	6.8	4.1 (2-14)
R-RSQ	53.0	14.1 (30-72)	47.8[a]	17.5 (25-74)	44.0	15.2 (24-70)
SDS	12.9	5.9 (4-21)	7.5[a]	6.1 (0-15)	5.8	5.5 (0-15)
SHAI Main	14.8	4.6 (8-20)	11.7	4.6 (4-17)	8.0	3.2 (1-12)
SHAI Negative consequences	3.4	2.7 (0-7)	2.3	2.2 (1-6)	2.9	1.9 (0-5)
SHAI-Avoidance	14.0	14.3 (0-42)	8.1	9.6 (0-22)	7.6	8.2 (0-19)
SHAI Reassurance	15.1	6.6 (6-24)	11.9	9.3 (1-23)	7.3	6.1 (0-17)

[a](*n*=8) *Note. n*=number of participants; SD=Standard Deviation; MADRS-S=Montgomery Åsberg Depression Rating Scale –Self Assessment; BAI=Beck Anxiety Inventory; PCL-C=The PTSD Checklist-Civilian Version; PSWQ=Penn State Worry Questionnaire; AAQ-II=Acceptance and Action Questionnaire-II; BIS=Body Image Scale; FAS=Fatigue Assessment Scale; PHQ-15=Public Health Questionnaire-15; R-RSQ=Rumination Scale of the Response Style Questionnaire; SDS=Sheehan Disability Scale; SHAI=The Short Health Anxiety Inventory

Table 4(on next page)

Number of individuals with complete data reaching reliable change from baseline to post-assessment and from baseline to follow-up assessment (*n*=7)

Note. n = number of participants; MADRS-S=Montgomery Åsberg Depression Rating Scale-Self Assessment; PCL-C=The PTSD Checklist-Civilian Version; BAI=Beck Anxiety Inventory. Reliable change scores were 7.7 for MADRS-S, 13.6 for PCL-C, and 22.9 for BAI.

Table 4:

Number of individuals with complete data reaching reliable change from baseline to post-assessment and from baseline to follow-up assessment (*n*=7).

	Baseline/Post-assessment	Baseline/Follow up assessment
MADRS-S	2	3
PCL-C	2	2
BAI	1	1
BAI, PCL-C and/or MADRS-S	3	4

Note. n = number of participants; MADRS-S=Montgomery Åsberg Depression Rating Scale-Self Assessment; PCL-C=The PTSD Checklist-Civilian Version; BAI=Beck Anxiety Inventory. Reliable change scores were 7.7 for MADRS-S, 13.6 for PCL-C, and 22.9 for BAI.

Table 5(on next page)

Summary of feasibility outcomes.

Table 5:
Summary of feasibility outcomes.

Outcome	Results
Recruitment	
Number identified via the Swedish Childhood Registry	*n*=286
Number of potential participants	*n*=213
Number contacted via telephone	*n*=111
Number contacted via post	*n*=98
Number of non-participating	
By exclusion	*n*=11
Declined participation	*n*=95
Lost to recruitment	*n*=94
Number assessed for eligibility	*n*=13
Number found ineligible	*n*=3
Number meeting inclusion criteria	*n*=10
Number included in the study	*n*=10
Reasons for non-participation	
Declined participation	**n=95**
No need for support	*n*=61
Online non-consent	*n*=10
Unable to travel to study sites	*n*=5
No interest prior to information given	*n*=4
No interest after receiving information	*n*=3
In counselling	*n*=3
Low trust in psychotherapy	*n*=2
No reason given	*n*=2
No perceived cancer	*n*=1
Intellectual disabilities	*n*=1
Feeling anger	*n*=1
Moved	*n*=1
Lack of time	*n*=1
Lost to recruitment	***n*=94**
No contact	*n*=91
Administrative failure	*n*=2
No address identified	*n*=1
By exclusion	***n*=14**
In psychological treatment	*n*=6
Age	*n*=2
In assessment for psychological treatment	*n*=3
Deceased	*n*=1
Unable to travel to study sites	*n*=1
Participation too burdensome	*n*=1
Attrition	10%

Percentage dropping out of the study Percentage dropping out of the treatment	10%
Data collection Percentage completing assessments Numbers of missing items	Post-treatment, 70% 3-month follow-up, 80% *n*=6
Treatment delivery Number of treatment sessions Length of treatment (weeks) Number of early treatment terminations Number of cancelled treatment sessions	Mean = 12.6, range = 7-15 Mean = 25, range = 18-36 *n*=2 Mean = 3.4, range = 0-11

4

Figure 1

Adapted CONSORT diagram illustrating the flow of participants through the study.

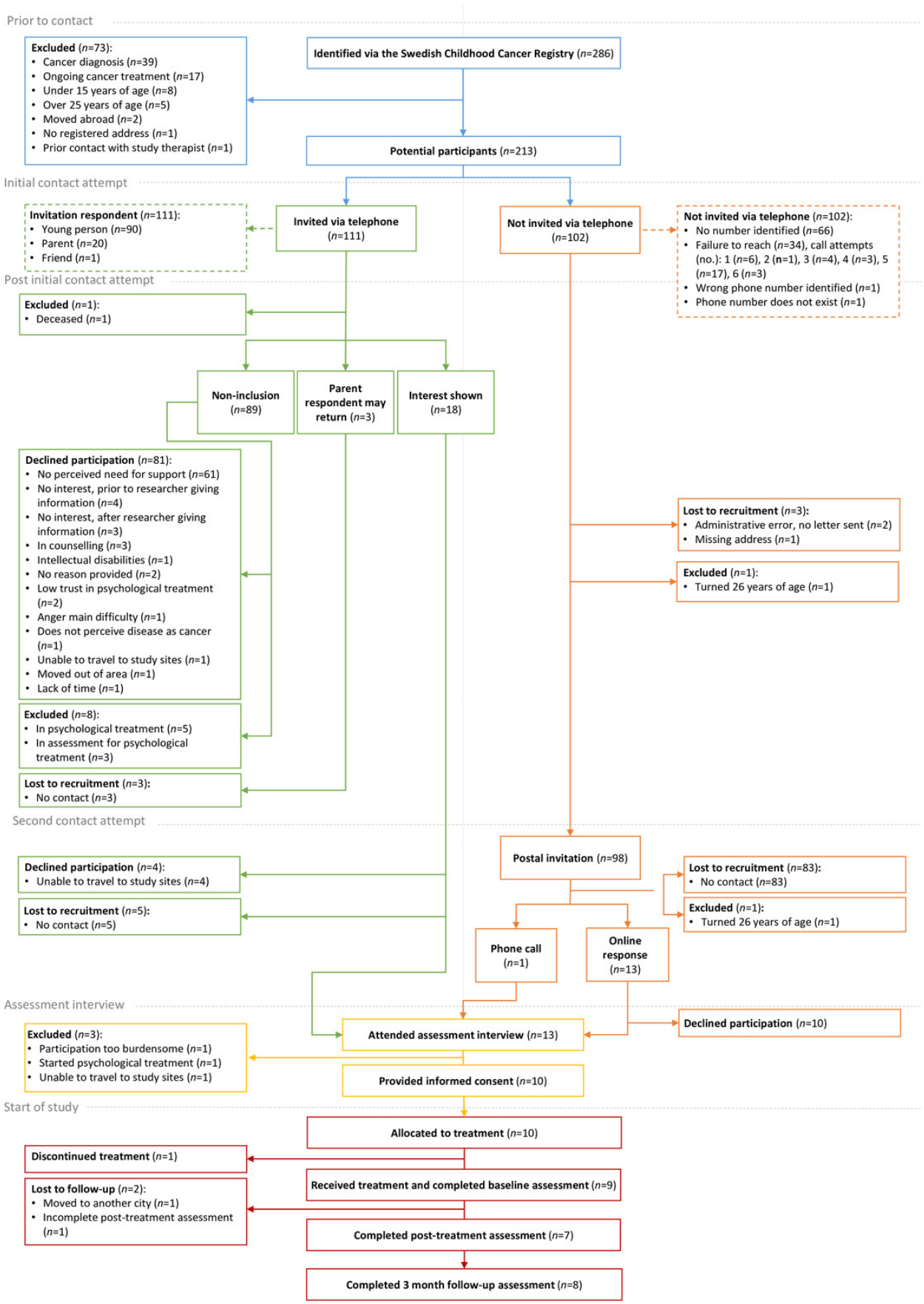

Prior to contact
Excluded (n=73):
• Cancer diagnosis (n=39)
• Ongoing cancer treatment (n=17)
• Under 15 years of age (n=8)
• Over 25 years of age (n=5)
• Moved abroad (n=2)
• No registered address (n=1)
• Prior contact with study therapist (n=1)
Identified via the Swedish Childhood Cancer Registry (n=286)
Potential participants (n=213)
Initial contact attempt
Invitation respondent (n=111):
• Young person (n=90)
• Parent (n=20)
• Friend (n=1)
Invited via telephone (n=111)
Not invited via telephone (n=102)
Not invited via telephone (n=102):
• No number identified (n=66)
• Failure to reach (n=34), call attempts (no.): 1 (n=6), 2 (n=1), 3 (n=4), 4 (n=3), 5 (n=17), 6 (n=3)
• Wrong phone number identified (n=1)
• Phone number does not exist (n=1)
Post initial contact attempt
Excluded (n=1):
• Deceased (n=1)
Non-inclusion (n=89)
Parent respondent may return (n=3)
Interest shown (n=18)
Declined participation (n=81):
• No perceived need for support (n=61)
• No interest, prior to researcher giving information (n=4)
• No interest, after researcher giving information (n=3)
• In counselling (n=3)
• Intellectual disabilities (n=1)
• No reason provided (n=2)
• Low trust in psychological treatment (n=2)
• Anger main difficulty (n=1)
• Does not perceive disease as cancer (n=1)
• Unable to travel to study sites (n=1)
• Moved out of area (n=1)
• Lack of time (n=1)
Excluded (n=8):
• In psychological treatment (n=5)
• In assessment for psychological treatment (n=3)
Lost to recruitment (n=3):
• No contact (n=3)
Lost to recruitment (n=3):
• Administrative error, no letter sent (n=2)
• Missing address (n=1)
Excluded (n=1):
• Turned 26 years of age (n=1)
Second contact attempt
Declined participation (n=4):
• Unable to travel to study sites (n=4)
Lost to recruitment (n=5):
• No contact (n=5)
Postal invitation (n=98)
Lost to recruitment (n=83):
• No contact (n=83)
Excluded (n=1):
• Turned 26 years of age (n=1)
Phone call (n=1)
Online response (n=13)
Assessment interview
Excluded (n=3):
• Participation too burdensome (n=1)
• Started psychological treatment (n=1)
• Unable to travel to study sites (n=1)
Attended assessment interview (n=13)
Declined participation (n=10)
Provided informed consent (n=10)
Start of study
Allocated to treatment (n=10)
Discontinued treatment (n=1)
Lost to follow-up (n=2):
• Moved to another city (n=1)
• Incomplete post-treatment assessment (n=1)
Received treatment and completed baseline assessment (n=9)
Completed post-treatment assessment (n=7)
Completed 3 month follow-up assessment (n=8)

Figure 2

Figure showing identified precipitatory and current cancer-related concerns and maintaining factors.

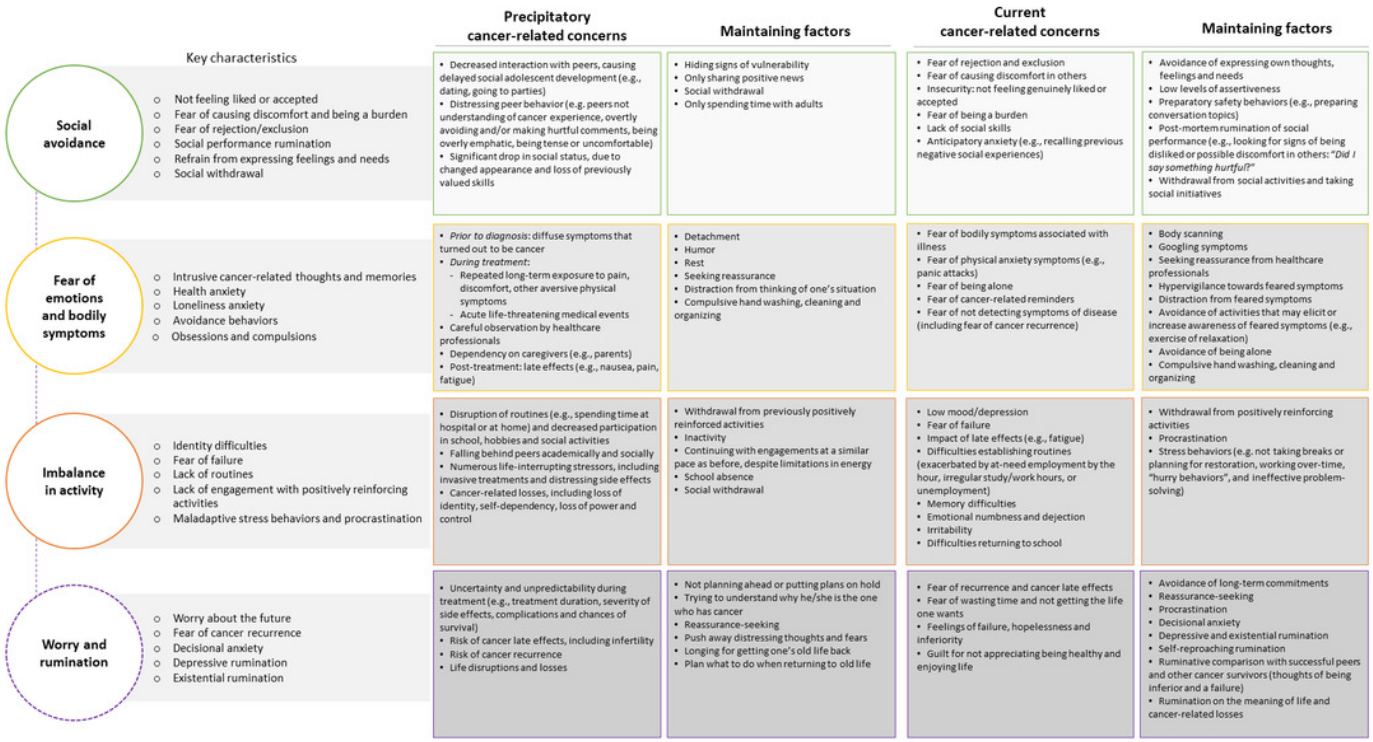
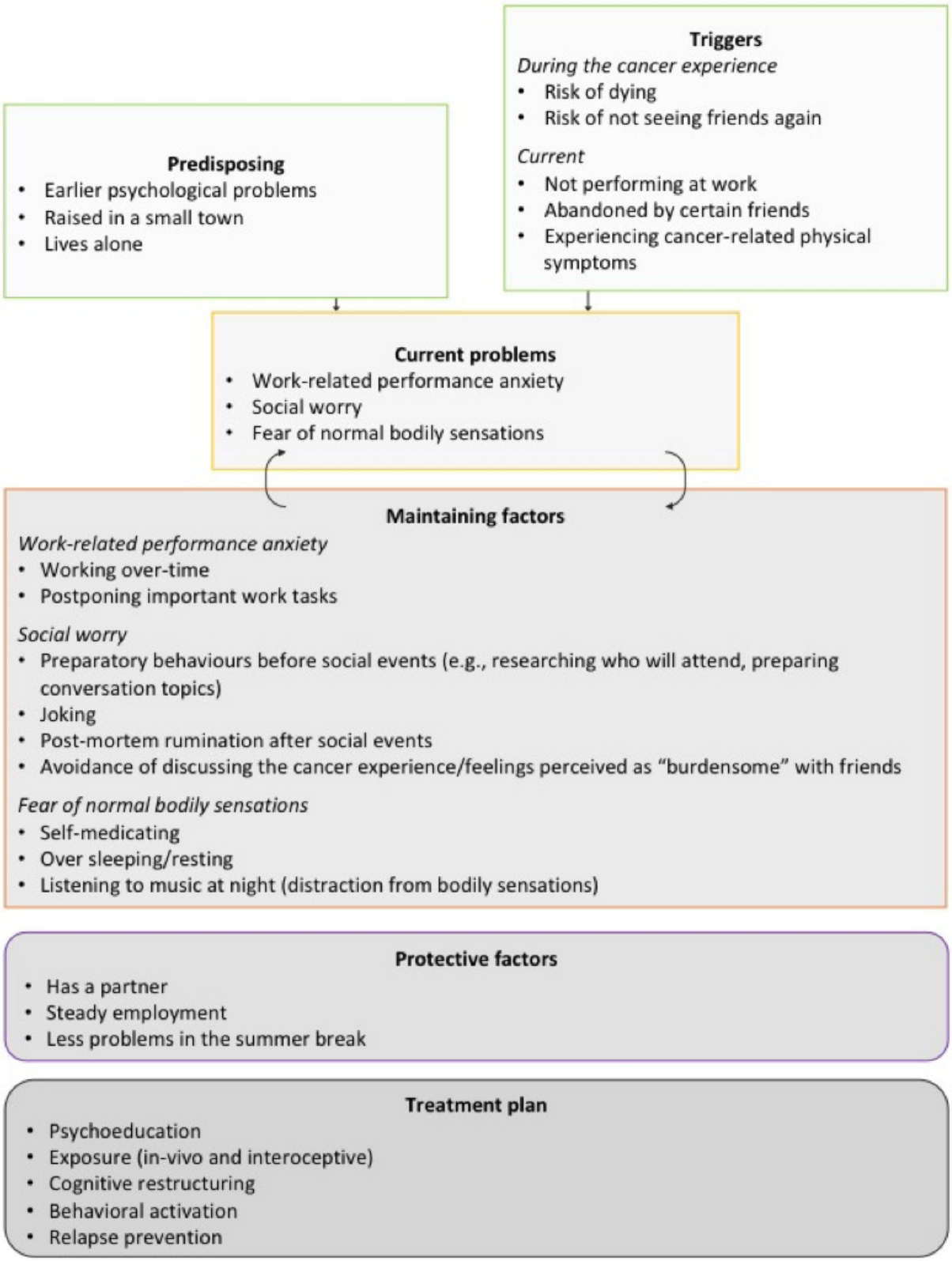


Figure 3

Figure showing identified precipitatory and current cancer-related concerns and maintaining factors.


Triggers
During the cancer experience
• Risk of dying
• Risk of not seeing friends again
Current
• Not performing at work
• Abandoned by certain friends
• Experiencing cancer-related physical symptoms
Predisposing
• Earlier psychological problems
• Raised in a small town
• Lives alone
Current problems
• Work-related performance anxiety
• Social worry
• Fear of normal bodily sensations
Maintaining factors
Work-related performance anxiety
• Working over-time
• Postponing important work tasks
Social worry
• Preparatory behaviours before social events (e.g., researching who will attend, preparing conversation topics)
• Joking
• Post-mortem rumination after social events
• Avoidance of discussing the cancer experience/feelings perceived as "burdensome" with friends
Fear of normal bodily sensations
• Self-medicating
• Over sleeping/resting
• Listening to music at night (distraction from bodily sensations)
Protective factors
• Has a partner
• Steady employment
• Less problems in the summer break
Treatment plan
• Psychoeducation
• Exposure (in-vivo and interoceptive)
• Cognitive restructuring
• Behavioral activation
• Relapse prevention