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Adolescent and young adult cancer survivors' perceptions of participating in a survey – Ethical and methodological considerations

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ABSTRACT

Purpose: The aim of this study was to understand patient-reported perception of participation in a population-based web-survey focusing on sensitive issues for adolescent and young adult cancer survivors.
Method: A population-based web survey for adolescent and young adult cancer survivors including a matched control group. Adolescent and young adult cancer survivors from the population-based Swedish National Cancer Registry from four of the six register holders at Regional Cancer Centers in Sweden. Controls were randomly identified from the Swedish National Population registry, from the same register holders.
Result: Of 729 eligible participants, 540 completed the survey i.e. 74% participation rate. The study population included 285 adolescent and young adult cancer survivors and 255 matched controls. None of the participants answered that the survey had a very negative impact on them and a minority of 43 (7.9%) of the 540 responded that they were mildly negatively affected by their participation in the study. There was a no significant difference between patients and controls regarding the negative effect of the participation ($p = 0.29$). Positive experiences of participating in the study were widely expressed and most participants (95%) found the study valuable.
Conclusions: These findings suggest that the benefits clearly outweigh the risks when adolescent and young adult cancer survivors participate in surveys including sensitive and trauma-related aspects, given that the study design is ethically sound and participants are approached carefully. We also present a modified ethical protocol for epidemiological surveys on adolescents and young adult cancer survivors.

1. Introduction

Adolescents and young adults (AYA) surviving cancer constitute a relatively new research population (Evan et al., 2006; Stevens, 2006; Wright et al., 2014; Zebrack et al., 2007). Patients themselves are a valuable source of information when evaluating their lives post cancer, for discovering suitable interventions and motivating proper care resources for the population. However, using a patient self-reporting questionnaire including questions on sexuality, depression and suicide can be challenging. Institutional ethical boards, national registries and boards on funding sometimes hesitate, motivating their concerns by arguing that the questions might be harmful or re-traumatizing for the participants. However, the risks attached to asking questions of a sensitive nature may be overestimated. Previous studies on asking sensitive questions on e.g. sexual issues or suicide showed by and large no significant negative impact on the participants (Dunberger et al., 2013; Eilegard et al., 2013; Evans et al., 2002; Harris and Goh, 2017; Omerov et al., 2014). In this specific population-based questionnaire, the aim has been to close the knowledge gap concerning the group of young individuals who have survived cancer during adolescence or young adulthood regarding: participation in surveys including sensitive and trauma-related questions, mental health, body-image, sexuality and fertility.

This paper focuses on variables in perception of participating in a population-based web survey. The remaining results have been or will be analyzed elsewhere (Olsson et al., 2018a, 2018b). A modified ethical protocol is also presented demonstrating a sensitive epidemiological approach when conducting web surveys on adolescent and young adult cancer survivors.

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Table 1
Adolescent and young adult cancer survivors and controls – a Swedish population-based survey: summary of ethical protocol for epidemiological surveys on adolescent and young adult cancer survivors.

I.	Preparation Carefully plan the inclusion criteria Same introductory letter to cancer survivors and controls To meet requirements of the Swedish law of secrecy, the researcher do not receive information about participants' cancer diagnoses. These were revealed by the informants themselves, after they had completed the survey. Make time; be prepared for long conversations with presumptive informants Create a database for all communication and contact information
II.	Introductory letter Researchers' contact information, e.g. telephone number, available 24 h Focus of the study and the questionnaire Option to end participation at any time without explanation Opportunity to decline contact or participation Several ways to decline contact or participation, e.g. telephone, text message, email and mail Information about coming telephone call – when and by whom Consideration and decision about how many letters to send at a time in order to be able to consider and respond to informants
III.	Telephone call Telephone call by trained interviewer Text message before first telephone call, providing information on what the call is about Careful sensitive "step-by-step" approach going from general questions to ones that are more detailed Being responsive and prepared for questions and need of support Providing support and help with referral if needed Encouraging contact again if help or support is needed Giving enough time for questions and support Accepting denial directly without further probing Repeating the option to end participation at any time without explanation Asking for consent to send login to questionnaire Obtaining participants' e-mail addresses for sending login to survey
IV.	During participation Continuity throughout the study with the same trained interviewers Interviewers being available and prepared for questions and support 24 h Providing support and help with referral if needed Giving enough time for questions and support Giving enough time for participation, e.g. being able to complete the survey online with a generous time allotment Sending an e-mail with a thank you note – and asking if there are any questions if no response Asking for consent to call again during participation – before calling again sending a text message informing about the call

With permission of author P. Omerov. The ethics of doing nothing. Suicide-bereavement and research: ethical and methodological considerations. Psychological Medicine (2014), 44, 3409–3420. A modification of the Summary of ethical protocol for epidemiological surveys on suicide-bereaved persons.

2. Methods

2.1. Study design, questionnaire development and participants

The study design was developed based on a stepwise method established by the research group at the Division of Clinical Cancer Epidemiology in Stockholm and Gothenburg, and used in more than 20 data collections focusing on cancer and survivorship (Bergmark et al., 1999; Dunberger et al., 2010; Eilegard and Kreicbergs, 2010). The threats to its validity were evaluated during the process by using an epidemiological method in the form of a hierarchical step-model for study design, analysis and data interpretation (Steineck et al., 2006). During the development, pre-study and main study, ethical considerations were applied and summarized in the modified ethical protocol using the sensitive approach presented in Table 1. The aim of the main study was to gather information on the physical and psychological health of adolescent and young adult cancer survivors that could be available for change and improvement in healthcare during and post cancer treatment. The hypotheses were based on clinical experience and previous interviews with young cancer survivors (Olsson et al., 2015). The questionnaire development included a literature review and collaboration with researchers from previous questionnaire developments, followed by 10 qualitative interviews with AYA cancer survivors to evaluate areas of concern and possible questions to include in the questionnaire. The study-specific questionnaire was developed and validated in several steps including comments from experts, researchers and clinicians. To validate the questionnaire we used face-to-face-validation with AYA cancer survivors, including patients with cognitive deficits, applying the with "thinking aloud" method to test their comprehension of the questions, to see if the main specific areas were covered and to test the length of the questionnaire (Hauge et al., 2015; van Leeuwen et al., 2015). The development and validation of the questionnaire and the pilot study development are presented in Fig. 1. The validation process also aimed to formulate the questions as inoffensively as possible. A revised draft was constructed and a pilot study conducted on 28 cancer survivors more than 5 years post cancer treatment. In the pilot study, the web-based questionnaire was tested concerning understanding of the study, the questionnaire and response rate. The overall participation rate in the pilot study was 71% and it resulted in minor refinement of the introduction letter to the questionnaire.

The questionnaire addressed to the cancer survivors contained 146 questions with follow-up questions and space for free comments. Every question was introduced by a short explanatory text. The first question was if they have had a cancer diagnosis or not, which directed them to further questions. The control group received a shorter questionnaire containing 95 questions, excluding questions on cancer treatment. Questions included demographics, and aspects such as education, occupation, social life, leisure activities, quality of life, wellbeing, psychosocial and mental health, self-image and sexuality, and questions on participation in the study.

The questions exclusively for the cancer survivors included questions about the cancer treatment period, including fertility. Examples of sensitive questions used in the survey: "Are you depressed?", "Have you had thoughts of suicide the past month?", "Have you had premature ejaculation the past month?" and "Have you experienced deep pain in your vagina during sexual activity the past six months?". Questions on participation included in the survey: "Do you think it is valuable to conduct this kind of survey?", "Do you think the survey has had a negative impact on you?", "Do you think the survey has had a positive effect on you?" and "Has the survey reminded you of events you had forgotten?" – to which the multiple-choice response alternatives were "Yes, very much", "Yes, moderately", "Yes, a little" or "No, not at all"; "Have any questions caused you distress?" – to which the responses were either "Yes" or "No", and if "Yes", then: "Which question/s?" Participants were also able to add free text. The response alternatives have been used in previous studies by the research group. (Dunberger et al., 2010; Skoogh et al., 2012). The questions on research participation were modified according to similar questions in previous research (Dunberger et al., 2013; Kreicbergs et al., 2005; Omerov et al., 2014; Skoogh et al., 2012).

The questionnaire was web-based and we used an on-line survey platform in collaboration with a health-care data company. This population-based study identified adolescent and young adult survivors of cancer treated for cancer at ages 15–29 during the period of 2010–2011, at least a year post treatment and alive at present. Participants were eligible for the study if they: 1/understood the Swedish language; 2/not were under treatment for cancer; 3/had a listed telephone number; and 4/were functionally/intellectually able to answer the questionnaire.

Names and addresses to cancer survivors were obtained from the population-based Swedish National Cancer Registry from four of the register holders, the north, west, southeast and central Regional Cancer Centers in Sweden. Controls were randomly identified from the Swedish

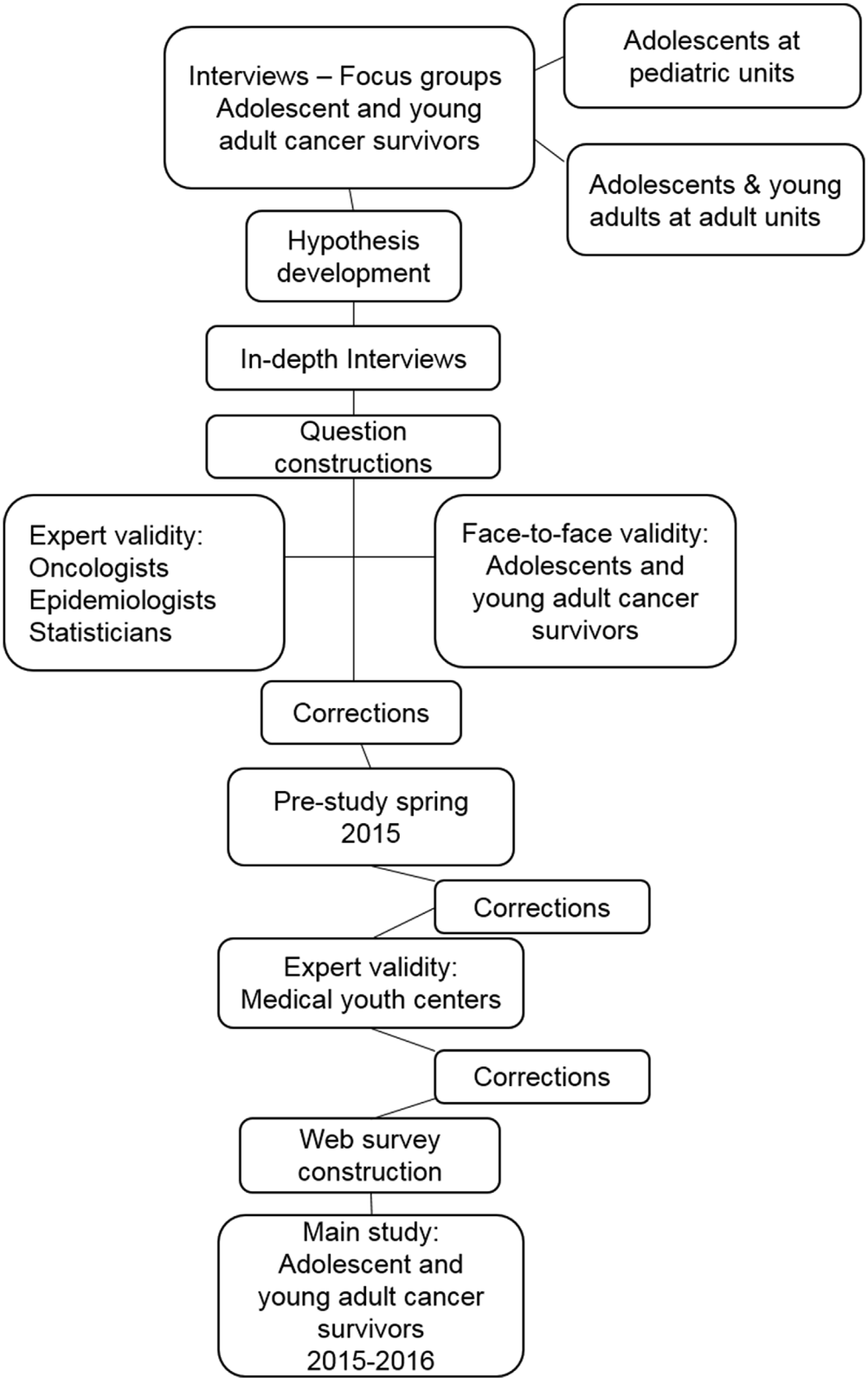

Fig. 1. Flow chart of questionnaire development.

National Population Registry, from the same register holders. The controls were matched by age, gender and place of residence. There was one matched control to each cancer survivor. The identification of the cancer survivors and the matching with the controls were carried out by the register holders at each Regional Cancer Center. The research group did not know whether the participants were cancer survivors or not until they had completed the questionnaire.

The Regional Ethical Review Board in Gothenburg, Sweden, granted ethical approval for this study with the reference number 691-13 and additional reference number 944-14.

2.2. Procedure

All potential participants were contacted via an introductory letter followed by a telephone call 1–2 weeks later. The procedure is described in Table 1 and Fig. 1. For ethical reasons it was made easy for them to decline participation before being contacted by telephone, by giving the research group's contact details – e-mail and mobile phone number. Fifty to seventy-five letters were sent at a time, enabling telephone contact to be made with all participants within 1–2 weeks. Data collection could thus be stopped if there were signs that the research was at all harmful to participants.

During the study, telephone contact was avoided on official holidays. Inclusion during the summertime was paused due to possible travelling and vacations. Prior to the first telephone call, a text-message was sent, to inform about the nature of the call, and about the study. In Sweden today, people receive many advertising calls and might be reluctant to answer an unknown number. This population was easily reachable during weekday afternoons between 4 and 8 pm and on Sunday afternoons. The phone calls were made by a research assistant or by the first author (M.O.), both being registered nurses with many years of clinical experience. Participants thus called were approached sensitively, starting with the question of whether they had received the letter about the study. A denial was accepted immediately without any persuasion to participate. Those giving informed oral consent to consider participation did so by giving the interviewer their e-mail address. Through an email, the participant received a personal username, password to the web questionnaire and information about an around-the-clock mobile phone number to the research team. The participants were reminded to complete the questionnaire 4 times in the following manner: the first reminder was a combined thank-you note and reminder sent by e-mail. The second reminder was made by a phone call and was also an opportunity for participants to ask questions if they had any. The third reminder was a text message and the fourth was an e-mail reminder.

The web-based questionnaire was returned to a database, each questionnaire with an identification number to maintain anonymity. A tailor-made database was used to enable a safe and systematic data collection, where all events and comments from participants were registered.

2.3. Statistical analyses

Statistical analyses were undertaken using SPSS version 23 and SAS 9.4 for Windows. Socio-demographic and descriptive characteristics were compared between the survivors and controls using the chi2-test for categorical variables. The non-parametric Kruskal-Wallis-test was used to test potential differences between groups with respect to positive and negative feelings on participating in a population-based survey. The level of significance was set at $p < 0.05$.

3. Results

3.1. Participants

From November 2014 to June 2016, 729 eligible cancer survivors and controls were included in the study. A total number of 540 participants completed the questionnaire, which gives a total response rate of 74%. For further information on participation, see Fig. 2. The cancer survivors had a lower education level than the controls ($P = 0.04$), otherwise there were no differences socio-demographically. Demographics and self-reported clinical characteristics are presented in Table 2.

3.2. Non-participants

Some of those contacted, 122 in number, declined participation. The main reason was that they were not interested in participation, with no other explanation given. A couple of participants declined due to mental health issues, and in two cases, relatives declined on the part of their relative. A few declined due to stress and lack of time. There is no information on to which category (cancer survivor or control) they belonged, since that question was asked in the survey, not on the phone. Six individuals declined because they did not want to think about cancer.

3.3. Agreed to participate but ended participation

Of the participants, 67 did not complete the survey. Most of the participants were grateful for the reminders in the form of e-mails, text messages on their mobiles and telephone calls since they claimed it was easy to forget the survey. Most participants did not give any explanation for not completing the survey. Some referred to “lack of time” and “having a complicated life at the moment”. None reported that the questions were too personal or sensitive.

3.4. Results of the completion experience

No participant responded that the survey had a very negative impact on her/him and only four out of the 540 (0.7%) answered that they were moderately negatively affected by participation in the study. There was a no significant difference between patients and controls regarding the negative effect of the participation. When combining moderate and little negative effect, 26 out of 273 (9.5%) of the cancer survivors reported some negative effect and 17 out of 246 (6.9%) of the controls did so. Parallel to this, positive experiences were widely expressed among most of the participating adolescent and young adult cancer survivors and most of the controls (98.8% totally) (Table 3). The adolescent and young adult cancer survivors found participation in this study more valuable than the controls ($p = 0.008$). On the question of whether participation had a positive effect, combining little/moderate/very positive responses, 184 out of 273 cancer survivors (67.3%) and 133 out of 246 controls (54%) reported a positive effect ($p < 0.01$).

A total of 49 participants out of 521 (9.4%) reported that questions in the survey caused them distress, 31 (10%) of cancer survivors and 18 (7%) of controls. Out of the 49 reporting distress, 30 made comments on which questions they felt caused distress (Table 4). Cancer survivors made 20 comments and controls 10. The two major questions cancer survivors reported as distressing were fertility and possible recurrence. Regarding questions on suicide, one of the cancer survivors reported them as distressing as did five of the controls. The cancer survivors stated that the survey reminded them of previous events in life to a higher degree than the controls ($p < 0.0001$). At the end of the survey, there was space for free comments and these comments mainly expressed gratitude for the opportunity to participate in order to help future patients. A few quotes from cancer survivors follow here: “I like it being an electronic survey since it is hard to talk about all topics in person.“, “You know that you are not alone.“, “My answers clearly show that I have to make changes in my life.“. In addition, a quote from the control group: “Depression and anxiety questions revealed that I might be going through something more serious than just temporary distress.“.

4. Discussion

We investigated how 540 adolescents and young adults perceived participating in a population-based web survey. A majority of the participants found participation in the survey valuable and only a minority felt that participation affected them negatively. A majority of the participants overall stated that participation affected them in a positive way. The adolescent and young adult cancer survivors found participation significantly more positive than the control group.

The findings correspond well with previous studies on surveys with

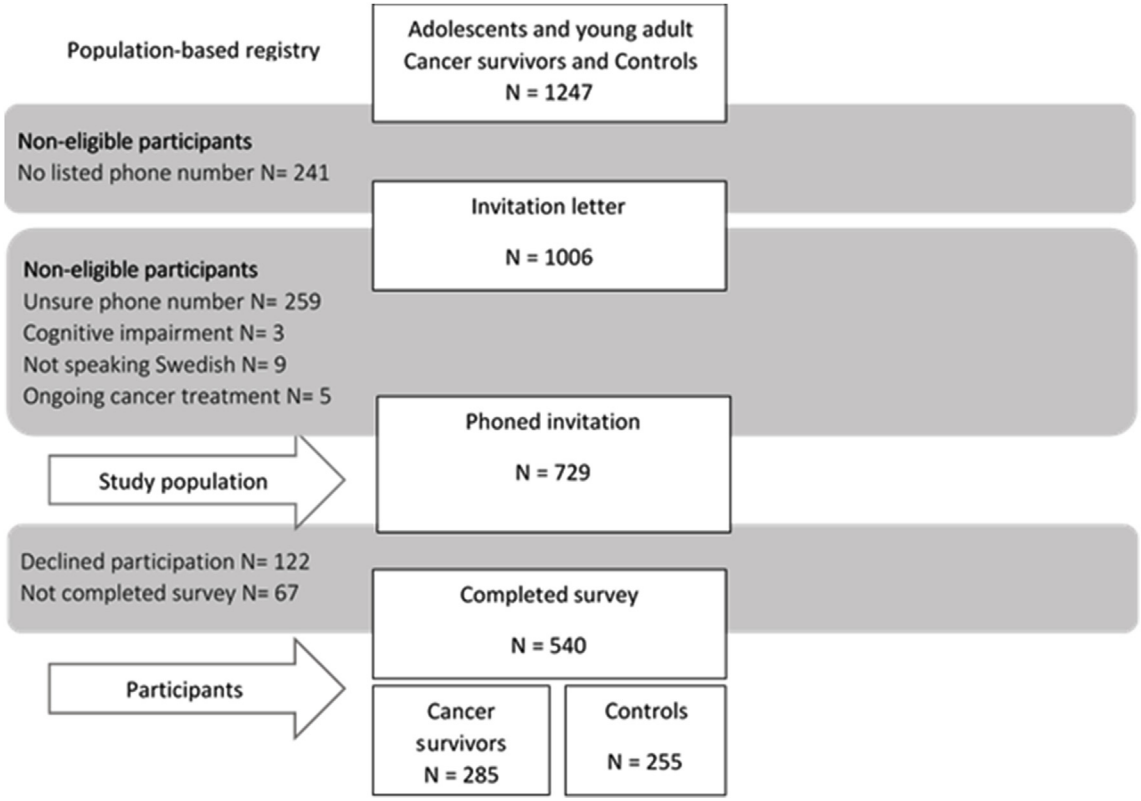

Fig. 2. Flow chart of participation.

sensitive questions on e.g. suicide, which have been shown to low negative impact on the participants (Dunberger et al., 2010; Evans et al., 2002; Harris and Goh, 2017; Kreicbergs et al., 2004; Omerov et al., 2014). There might be a negative emotional effect during the study, but despite this, participants evaluate participation as valuable (Beskow et al., 1990; Omerov et al., 2014). Negative emotional effects are described as: distress including feelings of depression; anxiety, embarrassment, and intrusion of privacy. Earlier studies have shown that these feelings quickly decrease after participation. There might be temporary distress when recalling traumatic events and this needs to be attended to during research (Jorm et al., 2007).

In this study, adolescent and young adult cancer survivors were involved at an early stage stating what areas they find important. In order to reduce the risk of misclassification, the questionnaire was validated face-to-face by adolescent and young adult cancer survivors with the same inclusion criteria as the study population. When planning and conducting research on sensitive or trauma-related events, it is important to have a sensitive approach and a strong research design including a control group (Jorm et al., 2007). The ethical protocol (Table 1) describes the ethical pathway throughout the study, to approach potential and current participants in a sensitive way. The introductory letter gave potential participants time to reflect on their decision before they were contacted. Before contacting the participants, a text message was sent before the actual telephone call, in order to let participants know that they were going to be contacted. This gave them the opportunity to decline contact and it gave the research team an opportunity to follow up with a question as to when the best time is to call. The personal phone call enabled the trained and experienced researcher calling to respond to participants' reactions to the contact, and it also gave potential participants the opportunity to ask additional questions on the study. Our impression is that potential participants did not have any problem in declining participation, which is congruent with findings in previous studies (Jorm et al., 2007). In addition to ethical principles on risks and benefits, there is an ethical concern regarding autonomy. In research discussions, the main concern is the potential opportunity for participants to decline participation. However, there are seldom discussions about participants' own capability of decision-making (Entwistle et al., 2010). Adolescents and young adult cancer patients have stated that they want to make the decision themselves, and not have it made by gate-keeping healthcare professionals (Taylor et al., 2016).

Participants did not contact the research team during the study due to temporary distress or need for assistance with referral to a counsellor. The around-the-clock number only received two calls during the study, one was about how to report working time and the other about how much information was wanted about the caller's sex life. The ethical protocol does not describe a pathway post participation, but if the participants needed to contact the research team, post participation, contact details were included in all e-mail and text message contacts.

Questions on depression and suicide have been the main concern for ethical boards and financial funders for this study, recommending exclusion of questions of that kind. However, for the adolescent and young adult cancer survivors in this study, the questions on fertility constituted the issue most frequently causing concern and distress. Despite this we do not consider this as an argument for excluding discussions on fertility. Rather, it highlights the importance of fertility issues for this age group, as frequently reported in previous studies (Benedict et al., 2016; Gorman et al., 2015). Other comments stated that questions in the survey normalized some of the physical and emotional conditions the participants were in and it let them know that they are not alone. The survey could benefit cancer survivors by shedding light on emotions and psychological states they might feel alone in and by creating a sense of coherence, as has earlier been stated (Odh et al., 2016). Additionally, being able to express feelings and experiences after a traumatic event by writing, may even be helpful for the individuals' psychological wellbeing (Kalantari et al., 2012; Smyth, 1998).

Table 2
Socio-demographics and self-reported clinical characteristics.

Characteristics	Survivors	Controls	*P value**
	N = 285 (52.8%)	N = 255 (47.2%)	
	n (%)	n (%)	
Age at study, years			
< 24	59 (21.5)	55 (22.0)	
25-30	124 (44.8)	108 (42.7)	
> 31	93 (33.7)	89 (35.2)	0.900
Age at diagnosis, years			
15-18	39 (13.7)		
19- 24	119 (41.8)		
25 -30	116 (40.7)		
Male	138 (48.4)	106 (41.6)	0.111
Female	147 (51.6)	149 (58.4)	
Marital status			
Married/cohabiting	152 (54.9)	148 (59.2)	0.606
In relationship	44 (15.9)	36 (14.4)	
Single	81 (29.2)	66 (26.4)	
Form of housing			
Parents	28 (10.1)	22 (8.8)	0.876
Independent living	248 (89.5)	227 (90.8)	
Other	1 (0.4)	1 (0.4)	
Living environment			
Rural area	51 (18.4)	38 (15.3)	0.371
Small town	173 (62.5)	162 (65.1)	
Larger city	53 (19.1)	47 (18.9)	
Education			
Elementary school	11 (4.0)	7 (2.8)	0.039
High school/College	154 (55.8)	116 (46.0)	
University/College	111 (40.2)	129 (51.2)	
Primary diagnosis (self-reported)			
Leukemia	17 (6.1)		
Lymphoma	39 (14.0)		
Testicular cancer	50 (17.9)		
Brain tumor	41 (14.7)		
Breast cancer	11 (3.9)		
Bone tumor	10 (3.6)		
Gynecological cancer	23 (8.2)		
Skin cancer	38 (13.6)		
Thyroid cancer	26 (9.3)		
Misc./Other	24 (6.0)		
Treatment (self-reported)			
Chemotherapy	122		
Radiation abdomen	24		
Other radiation	67		
Stem cell transplantation	19		
Surgery	221		

*The P-value was derived from a Chi2-test comparing young cancer survivors and controls.

Table 3
Participants' perceptions' of answering questionnaire.

	Survivors		Controls	
	N = 285 (%)	*P value*[a]	N = 255 (%)	*P value*[a]
Missing answers	12 (4.2)		9 (3.5)	
(97)Do you think it is valuable conduct such a survey?				
No, not at all	5 (1.8)		5 (2.0)	< 0.01
Yes, a little	42 (15.4)		44 (17.9)	
Yes, moderate	58 (21.1)		75 (30.5)	
Yes, very	171 (62.6)		122 (49.6)	
(98)Do you think the survey has had a negative impact in you?				
No, not at all	248 (90.5)		229 (93.1)	
Yes, a little	24 (8.7)		15 (6.1)	
Yes, moderate	2 (0.7)		2 (0.8) 0.46	
Yes, very	–		–	
(99)Do you think the survey has had a positive effect in you?				
No, not at all	91 (3.2)		113 (63.1)	< 0.001
Yes, a little	112 (45.3)		94 (38.1)	
Yes, moderate	54 (19.7)		31 (12.6)	
Yes, very	18 (6.6)		8 (3.3)	
(100) Has the survey reminded you of events you had forgotten?				
No, not at all	96 (35.0)		138 (56.1)	< 0.001
Yes, a little	109 (39.8)		90 (36.6)	
Yes, moderate	39 (14.2)		14 (5.7)	
Yes, very	31 (11.3)		4 (1.6)	

[a] The difference between young cancer survivors and controls analyzed by Kruskal-Wallis test.

Table 4
Questions Participants found affected them negatively.

	Survivors	Controls
	No.286(%)	No.255 (%)
Numbers participants giving free text comments	20 (6.9)	10 (3.9)
Fertility[a]	10 (50)	n.a.
Suicide	1 (5)	5 (50)
Recurrence[a]	5 (25)	n.a.
Body appearance	2 (10)	2 (20)
Depression	2 (10)	2 (20)
Sex	2 (10)	0 (0)
Physical activity	0 (0)	1 (10)
Counselling[a]	2 (10)	n.a.

Answers from free text condensed to question areas. Some participants wrote on multiple areas of questions.

[a] Questions on fertility, recurrence and counselling were not asked in the control group.

4.1. Strengths and limitations

This study has several strengths: firstly, having a population-based sample and matched controls, all identified through nationwide high-quality registers (Ludvigsson et al., 2009); and secondly, having a high participation rate for both cancer survivors and controls. The threats to validity were addressed using epidemiological methods as transferred to this study by hierarchical step-model (Steineck et al., 2006). Possible confounding factors were considered by matching the adolescent and young adult cancer survivors with the controls regarding socio-demographic variables. Controls had a higher education level than cancer survivors. An explanation for the higher education level might be that cancer survivors' education was delayed due to cancer treatment during their university years. A high participation rate, 74%, was attained in both the cancer survivors' and control groups. This rate should be high enough to avoid systematic errors related to misrepresentation.

The main limitation of this study concerns our not knowing all details why the non-participants, 26%, declined participation or did not complete the survey. Among those declining, the majority did not state any reason, though a few stated lack of time. For those not completing the survey, the main reason was lack of time. Nevertheless, many just did not want to continue their participation, without giving any reason. Whether or not this is due to the sensitive nature of the questions is unknown. We can only speculate that adolescents and young adults often have stressful lives. Some have small children, work full time or are studying for college and university exams, all of which interfere with participation in research. Another limitation is that the study design does not give us clinical information about non-participant patients. However, the distribution of the diagnoses among the

participating cancer survivors match the national distribution cancer diagnosed in this age-group. Furthermore, a limitation is that the survey only reached those understanding Swedish, which exclude non-Swedish speaking persons. Finally, this research group's method was to approach the participants as sensitively as possible. This approach was expensive and time-consuming which could be a potential limitation.

It is of greatest importance that ethical boards, research holders and research funders not hinder valuable research stating the research is too sensitive. There is a need for respect of the autonomy of potential participants' allowing them do decide for themselves whether to participate or not (General Assembly of the World Medical, 2014).

4.2. Conclusion

When making an ethical assessment of future studies, the scientific benefits must be carefully weighed against the possible burden laid on the shoulders of the participants. However, if the method that we have presented here is employed, our results suggest that this burden may be reduced to being very light or even non-existent, despite its addressing very personal and emotionally charged issues like sexual activity, suicide and death. To prevent or alleviate treatment-induced cancer-survivorship diseases we need information from survivors themselves and the data presented here shows that we should continue to do such research.

Conflict of interest statement

None declared.

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