The desire for children among adult survivors of childhood cancer: Psychometric evaluation of a cancer-specific questionnaire and relations with sociodemographic and psychological characteristics

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Abstract
Objective: Long-term childhood cancer survivors (CCS) are less likely to become parents than their peers of the same age. Previous research has suggested that besides fertility, this outcome is shaped by psychosocial factors such as emotional motives toward having a child. Drawing from a sample of CCS with survival times >25 years, we present the validation of a questionnaire assessing cancer-specific reproductive motives and concerns.

Methods: We evaluated the cancer-specific version of the Leipzig Questionnaire of Motives to have a Child (LKM-C) in a register-based sample of adult CCS (N = 632, 31% had children, 44.5% women). We conducted a confirmatory factor analysis and tested associations with sociodemographic characteristics and psychological symptoms (PHQ-9, GAD-2).

Results: The questionnaire showed good item discrimination parameters and reliability (α = 0.86). The two-factorial structure was confirmed with the independent scales “return to normalcy” and “illness-related worries (child’s/own health).” On average, CCS reported more motives in favor of a child than worries (P < .001; d = 1.12). Favorable attitudes were associated with the presence of a partnership and children, stronger current desire for a child, and fewer depressive symptoms. Worries were associated with an unfulfilled desire to have a child and elevated levels of depression and anxiety symptoms.

Conclusions: The LKM-C offers a brief measure of parenthood motivations in long-term CCS. Having a child signifies return to normalcy, health, fulfillment of life perspectives, and enrichment of the partnership. Less intense worries included illnesses of the child and concerns regarding one’s own health. The questionnaire could inform follow-up care, psychotherapy, and fertility treatments.
1 | INTRODUCTION

Following advances in diagnosis and treatment, 80% of children and adolescents diagnosed with cancer will become long-term survivors. However, physical, cognitive, and psychosocial late effects can diminish quality of life and disrupt psychosocial development including the start of a family. While negative effects of cancer and its treatment on fertility and pregnancy outcomes have recently come to the fore of clinical oncology, fewer studies have evaluated survivors' emotions concerning parenthood, or combined both aspects. In a recent American cohort study, 1067 childless, adult long-term childhood cancer survivors (CCS) (current age: M = 29.0(SD = 8.0) years) overestimated their personal risk of infertility. Thus, desires and concerns may also influence cancer survivors' reproductive intentions and well-being.

In this sense, the value-of-children-approach proposes a framework according to which decisions to have or not to have a child are the result of weighing up the reasons for and against it. For example, becoming a parent presents a possibility to live beyond one's own life span and to create something lasting. The benefits thus accrue to the individual as well as to the social unit of the couple or multigenerational family. On the other hand, having children is associated with economic and physical costs and time demands. One might also feel worry about facing a great and lasting responsibility for another human.

Previous research has indicated specific emotions, motives, and concerns related to parenthood after cancer. For instance, for 149 young male and female survivors (19-46 years) who had cancer at various sites, having children indicated the achievement of normalcy and hope for the future. On the other hand, the confrontation with their own mortality and fears of their own premature death might dissuade survivors from wanting children. Young CCS (N = 133, 16-24 years, >4 years after diagnosis) as well as a sample of 238 cancer survivors with a greater age span (18-45 years, diagnosed between the ages of 12 and 35) worried that their children could also be affected by cancer or by other diseases.

The recent adaptation of the Leipzig Questionnaire on Motives for Wanting Children for use in oncology (LKM-C, German title: "Leipziger Fragebogen zu Kinderwunschmotiven bei Krebs") stands in the tradition of the value-of-children approach as well as previous work and allows the quantitative assessment of various emotional motives and concerns influencing parenthood after cancer. It was previously evaluated in 175 young oncological patients (ages 18-39 years, survival times ≤5 years), but it has not yet been tested in adult CCS with longer survival times. However, an economical measure of such motives could be beneficial for the exploration of CCS' specific challenges and resources (i.e., in the context of follow-up care or psychotherapy).

The question of parenthood after cancer deserves attention as the overwhelming majority of cancer survivors have reported a wish to have a child as well as the need for more information about late effects regarding reproductive health. Importantly, reproductive concerns and unintentional childlessness were associated with mental distress in female cancer survivors.

We pursued three research aims:

1. Testing the LKM-C's psychometric characteristics in CCS >25 years after diagnosis
2. Exploring associations of different motives with sociodemographic aspects and mental distress
3. Investigating whether male and female CCS with an unfulfilled desire for children were more distressed than others

2 | METHODS

2.1 | Study design and participants

CCS were recruited in cooperation with the German Childhood Cancer Registry (GCCR). The nationwide GCCR systematically documents patients with childhood cancer residing in Germany since 1980. German CCS were eligible for participation if diagnosed with neoplasia according to the International Classification of Childhood Cancer (ICCC-3) between 1980 and 1990 before the age of 15, if registered at the GCCR, and if they had received antineoplastic treatment at one of 34 participating pediatric cancer centers. Survivors of Hodgkin lymphoma and a small group of former nephroblastoma patients could not be enrolled as they had taken part in other trials. A total of 2894 eligible survivors were invited to take part in the cross-sectional studies CVSS (Cardiac and Vascular late Sequelae in long-term Survivors of Childhood Cancer, clinicaltrials.gov-nr: NCT02181049) and PSYNA (Psychosocial long-term effects, health behavior and prevention among long-term survivors of cancer in childhood and adolescence). This invitation was accepted by 1002 CCS. Between 2013/09 and 2016/02, they were examined at the study center. We excluded 51 individuals due to subsequent malignant neoplasms, arriving at a baseline sample of 951 participants. A second assessment 1.5 to 2 years later consisted of a computer-assisted personal interview (CAPI) on health status and medical history, and a mailing of questionnaires concerning psychosocial aspects which included the LKM-C. As part of this second assessment, the LKM-C was completed by 632 individuals who constitute the main analysis set of this investigation. The study procedure, participants' diagnoses, and treatment-related information are described in more detail elsewhere.
CVSS and PSYNA are carried out in accordance with the ethical standards of the institutional research committee (approved by ethics review committee of Rhineland-Palatinate Chamber of Physicians, nr. 837.453.13(9138-F)) and with the Declaration of Helsinki. All participants gave written informed consent for study participation and data retrieval.

2.2 Materials and assessment

Disease-related data: CCS' illness- and treatment-related information was abstracted from primary health records of former treating medical centers and/or centrally documented individual therapy data available at the Society for Pediatric Oncology and Hematology's (GPOH) study centers. It was validated by trained medical staff.

Sociodemographic and psychological measures: Sociodemographic information was assessed via self-report as part of a computer-assisted personal interview (CAPI).

Cancer-specific motives for having a child were assessed using the Leipzig Questionnaire of motives to have a child for use in oncology (LKM-C). The development of its 20 items was based on previous research (detailed in a systematic review) and interviews with patients and professionals. It addresses specific motives related to having (survived) cancer such as survival motivation, hope for recovery, normalization of life, and worries about the child's health or the abilities to care for them. Participants rate each item on a Likert scale (0 = does not affect me to 4 = affects me strongly). For each of the LKM-C's two scales ("pro" = motives in favor of; "contra" = motives against having children), a sum score of 10 items can be calculated (range 0-40). In the validation sample, the LKM-C showed good item discrimination and reliability (α = 0.86). It also contains an additional item assessing participants' current desire to have children (1 = very weak to 7 = very strong). We further added a single question which concerned participants' (so far) unfulfilled desire for children (0 = no/1 = yes).

Socioeconomic status (SES) was defined according to Lampert and Kroll.26 The aggregated index ranges from 3 (lowest) to 21 (highest), based on the level of education, profession, and income.

We used the Patient Health Questionnaire (PHQ-9) to measure depression symptoms. Participants state the frequency of being bothered by each of the nine diagnostic criteria of major depression over the past 2 weeks (0 = not at all to 3 = nearly every day). The sum score of the PHQ-9 ranges from 0 to 27. In this investigation, it showed good internal consistency (α = 0.86). A sum score of ≥ 10 has achieved a sensitivity of 88% and a specificity of 88% for detecting major depression.27

Generalized anxiety symptoms were assessed with the two screening items (GAD-2) of the short form of the Generalized Anxiety Disorder Scale. Occurrence of "Feeling nervous, anxious or on edge" and "Not being able to stop or control worrying" over the last two weeks was rated on a Likert scale (0 = not at all to 3 = nearly every day). The GAD-2 has assessed generalized anxiety with good sensitivity (86%) and specificity (83%).28 It showed good internal consistency within the present study (ω = 0.78).

2.3 Statistical procedure

To pursue research aim 1, we calculated Cronbach’s α (separately for each scale) and carried out a confirmatory factor analysis (CFA) with two latent factors ("pro" and "contra"). We further calculated bivariate correlations and conducted a logistic regression analysis of parenthood status (children = 1; no children = 0) and a linear regression analysis of participants' current desire for children on their scores on the LKM-C's scales (controlling for sex and age). In the context of research aim 2, we calculated bivariate correlations and independent t-tests (for the binary variables sex and partnership). Research aim 3 was investigated by contrasting those who reported an unfulfilled desire to have children with the rest of the sample (using independent t-tests and χ²-tests).

P values correspond to two-tailed tests. In the case of univariate comparisons, they are supplemented by effect sizes (Cohen’s d). Test statistics of χ²-tests were used to calculate Cohen’s d.29 Analyses were performed using SPSS 23 for Windows.

3 RESULTS

3.1 Sample characteristics

Table 1 presents sociodemographic and disease-related and treatment-related information. In total, 632 participants completed the questionnaire. Leukemia survivors were the largest diagnosis group (42%). Participants' mean age at diagnosis was around 6 years. A current partnership was reported by 67% of the participants. Less than a third of the participants had children. Table 2 lists the sample's psychological characteristics. Participants rated motives in favor of children as higher than motives against having children (P < .001; d = 1.12). This was the case for men (P < .001, d = 1.22) and women (P < .001, d = 0.99). This was the largest effect size observed in the present study.

A nonresponder analysis (suppl. 1) showed slight differences between the 632 participants who had filled out the mailed questionnaire and the 319 participants who had participated in the study's first assessment, but had not returned the questionnaire. The latter were younger at the time of the study assessment and at diagnosis, had shorter follow-up times, lower SES, and a lower level of education. (All following analyses are based on the 632 responders.)

3.2 Psychometric characteristics of the LKM-C in the present sample

The scale range of 0 to 4 was fully exploited for each item, and there was sufficient variance. The corrected item-total correlations ranged between 0.357 and 0.699, indicating a good ability to differentiate (shown in Suppl. 2 which also lists English translations of all items). Internal consistency was α = 0.86 for each scale. The good results extended to different subgroups (participants with children: "pro": α = 0.84; "contra": α = 0.88; participants without children: "pro": α =
TABLE 1 Characteristics of study participants stratified by sex

|                      | All (N = 632) | Men (N = 351) | Women (N = 281) |
|----------------------|---------------|---------------|-----------------|
| **Demographics**     |               |               |                 |
| Age at first study assessment (SD)
  (y)                  | 34.92 (5.70)  | 35.42 (5.63)  | 34.29 (5.73)    |
| Socioeconomic status (SES)
  (SD)
  (y)                  | 13.26 (4.52)  | 13.82 (4.42)  | 12.57 (4.55)    |
| Unemployment (%)      | 68 (10.8)     | 21 (6.0)      | 47 (16.7)       |
| High school education (%) | 388 (61.4)    | 225 (64.1)    | 163 (58.0)      |
| Current Partnership (%) | 422 (66.8)    | 228 (65.0)    | 194 (69.0)      |
| Married (%)           | 233 (36.9)    | 131 (37.3)    | 102 (36.3)      |
| Children (%)          | 196 (31.0)    | 100 (28.5)    | 96 (34.2)       |
| 1 child (%)           | 85 (13.4)     | 40 (11.4)     | 45 (16.0)       |
| Several children (%)  | 111 (17.6)    | 60 (17.1)     | 51 (18.1)       |
| **Disease-related information** |           |               |                 |
| Age at diagnosis (SD)
  (y)                  | 6.34 (4.38)   | 6.69 (4.44)   | 5.90 (4.29)     |
| Follow-up time (SD)
  (y)                  | 28.07 (3.21)  | 28.22 (3.10)  | 27.88 (3.33)    |
| **Diagnosis (%)**     |               |               |                 |
| Leukemias             | 267 (42.2)    | 145 (41.3)    | 122 (43.4)      |
| CNS tumors            | 84 (13.3)     | 45 (12.8)     | 39 (13.9)       |
| Lymphomas             | 64 (10.1)     | 47 (13.4)     | 17 (6.0)        |
| Renal tumors          | 49 (7.8)      | 21 (6.0)      | 28 (10.0)       |
| Sympathic nervous system tumors | 48 (7.6) | 23 (6.6) | 25 (8.9) |
| Soft tissue sarcoma   | 51 (8.1)      | 31 (8.8)      | 20 (7.1)        |
| Malignant bone tumors | 34 (5.4)      | 18 (5.1)      | 16 (5.7)        |
| Germ cell tumors      | 18 (2.8)      | 9 (2.6)       | 9 (3.2)         |
| Retinoblastoma        | 8 (1.3)       | 6 (1.7)       | 2 (0.7)         |
| Hepatic tumors        | 4 (0.6)       | 2 (0.6)       | 2 (0.7)         |
| Carcinoma             | 4 (0.6)       | 3 (0.9)       | 1 (0.4)         |
| Others                | 1 (0.2)       | 1 (0.3)       | 0 (0)           |

Note.

*Sample range 24 to 49.

†Sample range 3 to 21 (sample range 3-21).

‡Sample range 0 to 14.98.

§Sample range 22.14 to 35.03.

0.87; “contra”: α = 0.81; men: “pro”: α = 0.85; “contra”: α = 0.86; women: “pro”: α = 0.88; “contra”: α = 0.86). We also tested the assumed two-factorial structure. The CFA model produced a statistically significant result (χ² = 983.2, df = 169, P > .001). The root mean square error of approximation (RMSEA = 0.07) and the comparative fit index (CFI = 0.91) suggested an acceptable to good fit of the model (criteria: CFI > 0.90; RMSEA < 0.10).35

There were strong positive correlations of the “pro”-scale with the current desire to have children (Table 3). The “contra”-scale did not correlate with the current desire to have children.

The logistic regression analysis of parenthood yielded a highly significant result (Nagelkerke R² = .461; P < .001). “Pro”-motives (OR = 1.08 (95%CI 1.05-1.10), P < .001) as well as “contra”-motives (OR = 0.97 (95%CI 0.95-0.99), P = .042) were statistically significant predictors. The linear regression analysis of the current desire for children also produced a highly significant result (adj. R² = 0.202, F (4, 506) = 32.103, P < .001). “Pro”-motives (β = 0.317, P < .001) as well as “contra”-motives (β = 0.03, P < .001) significantly contributed to the statistical prediction of the outcome.

3.3 | Associations of motives in favor of and against having a child with sociodemographic aspects and mental distress

The “contra”-scale was associated with higher levels of depression and anxiety symptoms, and lower socioeconomic status (Table 3). Participants in a partnership scored slightly higher on the “pro”-scale (M = 21.08 [SD = 9.94] vs M = 18.49 [SD = 8.88], P < .004, d = 0.27) and slightly lower on the “contra”-scale (M = 8.33 [SD = 7.45] vs M = 9.85 [SD = 9.77], P < .039, d = 0.17) than those who were single. There were no sex differences.

3.4 | CCS with an unfulfilled desire to have children

We investigated participants in more detail who reported an unfulfilled desire for children (OR = 1.10 (95%CI 1.05-1.10), P < .001) as well as “contra”-motives (OR = 0.97 (95%CI 0.95-0.99), P = .042) were statistically significant predictors. The linear regression analysis of the current desire for children also produced a highly significant result (adj. R² = 0.202, F (4, 506) = 32.103, P < .001). “Pro”-motives (β = 0.317, P < .001) as well as “contra”-motives (β = 0.03, P < .001) significantly contributed to the statistical prediction of the outcome.
the sample. These moderately sized differences pertained to male and female survivors.

4 | DISCUSSION

The present study extends knowledge of long-term CCS’ psychosocial situation with a systematic evaluation of emotional motives shaping their desire for children. Using the LKM-C, more than 600 adult CCS of both sexes were assessed concerning their positive motivations and concerns. Our analyses showed that the investigated questionnaire of both sexes were assessed concerning their positive motivations and concerns. Our analyses showed that the investigated questionnaire LKM-C has good psychometric properties and can be applied in long-term CCS. Further, its scales were related to relevant real-life outcomes.

The present assessment corroborated findings from qualitative and quantitative studies. Previous investigations using the Parenthood after cancer scale had also yielded quantitative estimations of concerns and positive parenthood motivation. In these studies, male and female cancer survivors generally expressed positive views of parenthood and many stated to feel even more appreciative of children after cancer, but they also reported great anxieties and uncertainties.

As there was no correlation of the “pro”- with the “contra”-scale in our study and the CFA confirmed a two-factor-solution, hopes and worries might represent unrelated, distinct parenthood motivations. Thus, survivors might feel that the positive aspects of having children apply irrespective of whether they consider having children is feasible for themselves, personally. This notion is supported by the finding that both “pro”- and “contra”-motives contributed to the statistical prediction of participants’ parenthood status and their current desire for children.

Along these lines, the greater standard deviation of the “contra”-scale’s sum score might reflect the comparatively greater diversity regarding CCS’ reproductive concerns.

The observed correlation patterns also correspond to the previous application of the LKM-C in young shorter term cancer survivors. These results speak for the usefulness of transferring the value-of-children approach into the psycho-oncological setting.

However, “contra”-motives might also represent the cognitive processing/mourning of an unfulfilled desire for children, or be based on cancer survivors’ medical history. Our finding that “contra”-scores were higher among CCS with an unfulfilled desire for children supports this assumption. Reproductive concerns (which included treatment-related late effects and other documentable reproductive characteristics) were also associated with unintentional childlessness in previous studies, e.g., using the Reproductive concerns after cancer scale. Hence, a more cautious labeling of the scales of the questionnaire could be “return to normalcy” (instead of “pro”-motives) and “illness-related worries (child/one’s own health)” (instead of “contra”-motives).

Lastly, our results indicate that cancer survivors with an unfulfilled desire to have children might be a particularly vulnerable group, corroborating previous research. The large age range of our sample might imply certain heterogeneity of cases within this group. For those of younger age, actual family planning might not have started yet, whereas older participants potentially have had a history of trying and disappointment. Correspondingly, previous research has found that cancer survivors struggle with reproductive concerns irrespective of their age, previous efforts to become pregnant, and even already existing children. Thus, having children as well as remaining childless appear to be crucial issues over the entire life span of cancer survivors of both sexes.

4.1 | Clinical implications

The present results suggest that the LKM-C could be a useful instrument in the context of CCS’ standard care as well as counseling. It fulfills the need for an economical assessment of worries, resources, and wishes. It also gives insights into which issues are particularly important for the respective respondent. As the relatively small effect sizes (observed in the context of regression analyses as well as univariate comparisons) caution against the assumption that the contents of LKM-C might be of clinical relevance to all CCS, we suggest the questionnaire’s use as a supportive tool which could facilitate conversations aimed at determining survivors’ most important concerns.

### TABLE 3 Bivariate correlations of the scales of the LKM-C with other measures of interest

|              | "Pro" - Motives | "Pro" - Motives | Current Desire for Children | Depression Symptoms | Anxiety Symptoms | Age | Socioeconomic Status |
|--------------|-----------------|-----------------|-----------------------------|---------------------|-----------------|-----|----------------------|
| "Pro" - motives | 0.084           | 0.317**         | -0.134**                   | -0.075              | 0.036           | 0.024 |
| "Contra" - motives | -0.003          | 0.443**         | 0.404**                    | 0.031               | -0.200**        |     |
| Current desire for children |               | -0.067          | 0.751**                    | 0.067               | -0.262**        |     |
| Depression symptoms |               |                 |                             |                     |                 |     |
| Anxiety symptoms |               |                 |                             |                     |                 |     |
| Age            |                 |                 |                             |                     |                 | 0.064 |
| Socioeconomic status |           |                 |                             |                     |                 |     |

Note
*P ≤ .05
**P ≤ .01, two-tailed.
Perhaps, positive valuations of having children after cancer (eg, focusing on the positive, achieving normalcy) could inform psychological interventions as they capture powerful emotional motives. Unfortunately, the uncertainty regarding the accuracy of participants’ illness-related worries mirrors their lived experience: It has been noted that cancer survivors’ assumptions regarding their fertility as well as potential health risks are based on subjective beliefs rather than medical tests and might be overstated. Participants in our study gave reasons why they did not wish to undergo fertility testing, eg, to avoid assignments of blame. Financial barriers might also play a role.

However, as the majority of cancer survivors have expressed a wish for having children, failing to address issues of fertility and concerns about parenthood might implicate uncertainties, distress, and missed opportunities. Importantly, cancer survivors of both sexes have stated that infertility strained their romantic relationships. Thus, CCS should be supported in decisions around fertility testing and treatments, alternative approaches such as surrogacy or adoption, and also in processing grief and disappointment. More research is needed to investigate emotional aspects of parenthood after cancer in men and women, ideally in conjunction with fertility. In the context of follow-up care, accessible offers of such services would allow for an evidence-based individual consultation which could address biological realities as well as wishes and concerns in a sensible way.

4.2 | Limitations

The results of the present study need to be interpreted in light of its limitations. There are a number of reasons why CCS who took part in the study might not be representative for the total population of CCS. First, Hodgkin lymphoma as well as some neuroblastoma survivors could not be enrolled. Second, regarding the entire invited CCS cohort, only 22% filled out the LKM-C. As we lack data on CCS who did not accept the study invitation, we are unable to empirically investigate their characteristics. Within our study, a nonresponder analysis indicated that our results might especially apply to CCS with higher SES, higher current age, and higher age at diagnosis, although group differences were small. Third, the relevance of cancer-specific motives might depend on survivors’ time since diagnosis. For instance, concerns might be more salient for those still undergoing treatment. Lastly, current and future survivors’ situation might not be comparable to our participants’ experiences (having been diagnosed between 1980 and 1990). There have been great advances in the knowledge about the effects of cancer and its treatment on reproductive health as well as options for fertility preservation. Furthermore, family policy as well as societies’ attitudes toward children form a societal framework which changes over time.

The cross-sectional study design cautions against assumptions regarding the direction of the observed effects, eg, associations of the LKM-C’s scores and the presence of children. While we assume that emotional motives and concerns influence the desire for children, having children could also influence how participants feel and think about the questionnaire’s topics.

Another limitation is the absence of information about participants’ fertility status and whether they had undergone testing. Thus, it was not possible to ascertain to what extent the concerns expressed in the questionnaire had a biological/functional basis. The reports of an unfulfilled desire for children suggest that such difficulties could affect at least a subgroup of participants. However, we do not know the individual reasons for these statements and for the relatively few participants with children (which corresponds to previous findings). While our sample comprised many leukemia survivors among which infertility is a common late effect, CCS in general have experienced delays in achieving social and psychosexual milestones.

5 | CONCLUSION

We report the first use of the LKM-C within a unique sample of CCS with survival times >25 years. The scale shows good psychometric properties and reliability. The relations with participants’ parenthood status and current desire for children attest to its validity. The LKM-C is a useful instrument to explore incentives and challenges surrounding parenthood in long-term CCS. Its use could inform long-term follow-up care as well as psychosocial support. Our study indicates that reproductive concerns could be a previously understudied risk factor for CCS’ mental health.

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CONFLICT OF INTEREST

None.

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DATA AVAILABILITY STATEMENT
The written informed consent of the study participants is not suitable for public access to the data, and this concept was not approved by the local data protection officer and ethics committee. Access to data at the local database in accordance with the ethics vote is offered upon request at any time. Interested researchers make their requests to the Principal Investigators of the CVSS/PSYNA study (philipp.wild@unimedizin-mainz.de).

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Additional supporting information may be found online in the Supporting Information section at the end of the article.