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ORIGINAL ARTICLE

Impaired social functioning in adolescent and young adult sarcoma survivors: Prevalence and risk factors

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Abstract

Background: Sarcomas account for almost 11% of all cancers in adolescents and young adults (AYAs; 18–39 years). AYAs are increasingly recognized as a distinct oncological age group with its own psychosocial challenges and biological characteristics. Social functioning has been shown to be one of the most severely affected domains of health-related quality of life in AYA cancer survivors. This study aims to identify AYA sarcoma survivors with impaired social functioning (ISF) and determine clinical and psychosocial factors associated with ISF.

Methods: AYAs from the population-based cross-sectional sarcoma survivorship study (SURVSARC) were included (n = 176). ISF was determined according to the European Organization for Research and Treatment of Cancer Quality of Life Questionnaire Core 30 social functioning scale, and age- and sex-matched norm data were used as reference.

Results: The median time since diagnosis was 6.2 years (range, 1.8–11.2). More than one-quarter (28%) of AYA sarcoma survivors experienced ISF. Older age, higher tumor stage, comorbidities, lower experienced social support, uncertainty in relationships, feeling less attractive, sexual inactivity, unemployment, and financial difficulties were associated with ISF. In a multivariable analysis, unemployment (OR, 3.719; 95% CI, 1.261–10.967) and having to make lifestyle changes because of financial problems caused by one's physical condition or medical treatment (OR, 3.394; 95% CI, 1.118–10.300) were associated with ISF; better experienced social support was associated with non-ISF (OR, 0.739; 95% CI, 0.570–0.957).

Conclusion: More than one-quarter of AYA sarcoma survivors experience ISF long after diagnosis. These results emphasize the importance of follow-up care that is not only disease-oriented but also focuses on the psychological and social domains.

The first two authors contributed equally and share first authorship.

Plain Language Summary

- Sarcomas account for almost 11% of all cancers in adolescents and young adults (AYAs; 18–39 years). The AYA group is increasingly recognized as a distinct oncological age group with its own psychosocial challenges and biological characteristics.
- Social functioning has been shown to be severely affected in AYA cancer survivors.
- A population-based questionnaire study to identify AYA sarcoma survivors with impaired social functioning (ISF) and determine factors associated with ISF was conducted. More than one-quarter of AYA sarcoma survivors experience ISF long after diagnosis. These results emphasize the importance of follow-up care that is not only disease-orientated but also focuses on the psychological and social domains.

KEYWORDS
adolescents and young adults, health-related quality of life, sarcoma, social functioning, survivorship

INTRODUCTION

Sarcomas are rare malignancies of the bone and soft tissue that originate from mesenchymal stem cells. With more than 70 histological subtypes, sarcomas are very heterogeneous tumors.[1] This heterogeneity of soft tissue sarcomas and bone sarcomas further extends into the great variation in morphology, age of occurrence, tumor localization, and clinical behavior.[2] Together, sarcomas have an incidence of approximately 5 cases per 100,000 people per year, thus meeting the formal criteria for rare tumors.[3] The 5-year overall survival is approximately 53% and 66% for soft tissue sarcomas and bone sarcomas, respectively.[4]

Sarcomas demonstrate a diverse nature across the age spectrum in terms of both incidence and histological subtype. Although they represent only 1% of all adult cancers, they comprise 11% of all cancers in people younger than aged 29 years.[5,6] Additionally, the challenges that cancer patients face are highly variant at different ages. One age group that has been increasingly recognized as an independent oncological age group with its own psychosocial challenges and biological characteristics are adolescents and young adults (AYAs; aged 18–39 years at diagnosis). AYAs are diagnosed at an emotionally, cognitively, and socially challenging time in their lives.[7] Something as far-reaching as a cancer diagnosis at this age could interfere with the acquisition of regular developmental milestones, such as graduating, reaching financial independence, and establishing relationships.[8] As a result, AYAs diagnosed with cancer report lower scores on Health-Related Quality of Life (HRQOL) scale, including a negative impact on financial life, body image,[9] sexual life and intimate relationships,[9,10] and feelings of social isolation.[8]

A recent study on HRQOL in patients with sarcoma showed that AYA sarcoma survivors report statistically significant and clinically relevant lower social functioning scores compared with their age-matched normative population.[11] This corresponds to previous research reporting social functioning as one of the most severely affected domains of HRQOL in AYA cancer survivors.[12–14] Social functioning in the AYA population can be defined as a domain of HRQOL, focused on limitations and difficulties with participation in social activities and functioning in relationships. Social functioning may be influenced by several factors, including employment status, financial burden, body image, and social support.[15]

The rarity of sarcomas and the intensive treatment they require in combination with the social vulnerability of AYAs, gives AYA sarcoma survivors a high risk of impaired social functioning (ISF). In-depth data on social functioning of AYA sarcoma survivors are scarce; little is known about the challenges that AYA survivors experience concerning social functioning, and risk factors for ISF have not been adequately researched. Therefore, this study aimed to (1) identify AYA sarcoma survivors with ISF and (2) determine socio-demographic, tumor, treatment, and psychosocial characteristics associated with ISF.

METHODS

Study design and participants

Data from the sarcoma survivorship study (SURVSARC) were used. This is a population-based, cross-sectional cohort study aimed to assess various aspects related to HRQOL among adult (≥18 years) sarcoma survivors, registered in the Netherlands Cancer Registry (NCR). All sarcoma patients diagnosed between January 1, 2008, and

December 31, 2016, at one of the six participating Dutch sarcoma expertise centers was eligible for inclusion. In this study, only AYA patients (aged 18–39 years at diagnosis) who completed the social functioning questions from the questionnaire were selected from the SURVSARC study. More detailed information on the study design has been published previously.[11]

Data collection

Participants were invited by their treating physician to complete the questionnaire between October 2018 and June 2019 within the Patient Reported Outcomes Following Initial treatment and Long-term Evaluation of Survivorship data management system. Time since diagnosis at the time of completing the questionnaire varied between 2 and 10 years.

Study measures

Sociodemographic, tumor, and treatment characteristics

Sociodemographic characteristics were partly obtained from the NCR (age and sex) and partly self-reported by the respondents (employment status and relationship status). Tumor and treatment characteristics were obtained from the NCR.

Social functioning and experienced social support

The social functioning scale of the European Organization for Research and Treatment of Cancer Quality of Life Questionnaire Core 30 (EORTC-QLQ-C30) was used to determine if survivors reported ISF or non-ISF. According to the evidence-based guidelines for interpreting the EORTC-QLQ-C30, the cutoff for a medium or large clinically relevant difference is 11 points,[16] which we used as a cutoff. As a reference score, we used the average social functioning score of the age- and sex-matched norm population, which was 94, and has been reported previously for this exact study population.[11] Additionally, the more exhaustive EORTC-Computer Adaptive Test Social Functioning questionnaire was completed, which consists of 13 items. Experienced social support was assessed on a scale from 0 to 10, with an item originating from the Quality of Life for Cancer Survivors questionnaire.[17]

Relationships, sexual functioning, and body image

To assess relationship concerns, eight items from the Impact of Cancer, version 2, were used.[18] Two items on sexual health and three items on body image originating from the EORTC item library were used.[19]

Workability and financial toxicity

Survivors were asked if they had a paid job at the time of filling out the questionnaire. One additional item from the Workability Index questionnaire was used that asked about survivors' capability of doing their job compared with before their illness on a scale from 0 to 10.[20] Nine items on financial toxicity from the EORTC item library were used to assess financial toxicity.[19]

Statistical analyses

Comparative analyses between the ISF group and non-ISF group on sociodemographic, tumor, treatment, and psychosocial characteristics were conducted. χ^2 tests and *t*-tests were used for the analysis of categorical and continuous variables, respectively.

For statistical analyses, the five answering options (strongly disagree, disagree, neutral, agree, strongly agree) assessing relationship concerns were brought back to three options (disagree, neutral, agree). The 4-point Likert scale of all EORTC items was dichotomized: "not at all" was altered to "disagree" and "a little," "quite a bit,"' and "very much" were grouped into "agree." The rationale for this was to improve the clinical interpretability and increase the statistical power of the analyses.

The variables included in the multivariable analysis were those that were statistically significant in the univariate analyses ($p < .05$). Because of the limited number of patients, we could not include all those variables in the multivariable analysis. When more than one item was statistically significant within one concept (e.g., sexual functioning), the item that was most strongly associated with ISF in the univariate analysis (highest odds ratio) was selected (Table S1). There was no multicollinearity between the variables included in the multivariable analysis.

All statistical analyses were performed using SPSS Statistics (IBM Corporation, version 27.0, Armonk, NY) and p values $< .05$ were considered statistically significant.

RESULTS

Sociodemographic, tumor, and treatment characteristics

In total, 186 AYA survivors were included in the SURVSARC study, of whom 176 completed the social functioning scale of the EORTC-QLQ-C30 and were included in this secondary analysis. The only statistically significant difference in terms of sociodemographic, tumor, and treatment characteristics between the 10 excluded survivors and the 176 included survivors was that the excluded survivors more often had no education or only primary/secondary school education (6.3% vs 30%, $p = .030$). A responder vs nonresponder analysis for the entire population of the SURVSARC study has been published previously.[21] Of the included survivors, 45% were male and

TABLE 1 Sociodemographic, tumor, and treatment characteristics.

Sociodemographic, tumor and treatment characteristics	Total *N* = 176 (100)	Nonimpaired SF *N* = 127 (100)	Impaired SF *N* = 49 (100)	*p*
Sex				
Male	79 (44.9)	55 (43.3)	24 (49.0)	.498
Female	97 (55.1)	72 (56.7)	25 (51.0)	
Age at diagnosis, years				
Median (range)	30 (18–39)	28 (18–39)	32 (18–39)	.043
Mean (±SD)	29.4 (6.6)	28.8 (6.8)	31.1 (5.8)	
Time since diagnosis, months				
Median (range)	74.50 (21–135)	73 (22–132)	77 (21–135)	.489
Mean (±SD)	76.0 (31.4)	75.0 (31.2)	78.7 (32.2)	
Educational level				
No education, primary school, or secondary school	11 (6.3)	6 (4.7)	5 (10.2)	.334[a]
Vocational qualification	73 (41.5)	52 (40.9)	21 (42.9)	
College or university	92 (52.3)	69 (54.3)	23 (46.9)	
Comorbidities				
None	102 (58.0)	80 (63.0)	22 (44.9)	**.022**[a]
One	57 (32.4)	39 (30.7)	18 (36.7)	
Two or more	17 (9.7)	8 (6.3)	9 (18.4)	
Histology				
STS	102 (58.0)	78 (61.4)	24 (49.0)	.134 (STS vs BS)
DFSP	24 (13.6)	22 (17.3)	2 (4.1)	
Liposarcoma	22 (12.5)	16 (12.6)	6 (12.2)	
Myxofibrosarcoma	4 (2.3)	2 (2.4)	1 (2.0)	
Leiomyosarcoma	7 (4.0)	4 (3.1)	3 (6.1)	
Rhabdomyosarcoma	6 (3.4)	5 (3.9)	1 (2.0)	
MPNST	9 (5.1)	6 (4.7)	3 (6.1)	
Synovial sarcoma	10 (5.7)	8 (6.3)	2 (4.1)	
Vascular sarcoma	2 (1.1)	1 (0.8)	1 (2.0)	
Other STS	18 (10.2)	13 (10.2)	5 (10.2)	
BS	74 (42.0)	49 (38.6)	25 (51.0)	
Osteosarcoma	29 (16.5)	22 (17.3)	7 (14.3)	
Chondrosarcoma	24 (13.6)	11 (8.7)	13 (26.5)	
Chordoma	1 (0.6)	0 (0.0)	1 (2.0)	
Ewing sarcoma	16 (9.1)	13 (10.2)	3 (6.1)	
Other BS	4 (2.3)	3 (2.4)	1 (2.0)	
Grade				
Low	99 (58.9)	72 (58.1)	27 (61.4)	.702
High	69 (41.1)	52 (41.9)	17 (38.6)	
Missing	8	3	5	
Stage				
I	89 (53.6)	73 (60.3)	16 (32.7)	.026[a]

TABLE 1 (Continued)

Sociodemographic, tumor and treatment characteristics	Total N = 176 (100)	Nonimpaired SF N = 127 (100)	Impaired SF N = 49 (100)	p
II	50 (30.1)	31 (25.6)	19 (38.8)	
III	9 (5.4)	5 (4.1)	4 (8.2)	
IV	18 (10.6)	12 (9.9)	6 (12.2)	
Missing	10	6	4	
Localization				
Extremity	86 (46.2)	56 (44.1)	25 (51.0)	.233[a]
Upper extremity	18 (10.2)	11 (8.7)	7 (14.3)	
Lower extremity	63 (35.8)	45 (35.4)	18 (36.7)	
Nonextremity	100 (53.8)	71 (55.9)	24 (49.0)	
Head and neck	14 (8.0)	9 (7.1)	5 (10.2)	
Thoracic	17 (9.7)	13 (10.2)	4 (8.2)	
Abdomen	10 (5.7)	9 (7.1)	1 (2.0)	
Breast	2 (1.1)	1 (0.8)	1 (2.0)	
Skin	25 (14.2)	22 (17.3)	3 (6.1)	
Pelvis	12 (6.8)	6 (4.7)	6 (12.2)	
Other	15 (8.5)	11 (8.7)	4 (8.2)	
Treatment				
Surgery	69 (39.2)	55 (43.3)	14 (28.6)	.336[a]
RT	2 (1.1)	1 (0.8)	1 (2.0)	
CT	1 (0.6)	1 (0.8)	0 (0.0)	
Surgery and RT	44 (25.0)	27 (21.3)	17 (34.7)	
Surgery and CT	35 (19.9)	25 (19.7)	10 (20.4)	
Surgery, RT, and CT	20 (11.4)	15 (11.8)	5 (10.2)	
RT and CT	5 (2.8)	3 (2.4)	2 (4.1)	

Abbreviations: BS, bone sarcoma; CT, chemotherapy; DFSP, dermatofibrosarcoma protuberans; MPNST, malignant peripheral nerve sheath tumor; RT, radiotherapy; STS, soft tissue sarcoma.

[a]Fisher's exact.

the median age at diagnosis was 30 years (Table 1). Survivors had a median time since diagnosis of 6.2 years.

More than one-half of survivors with ISF (55%) had one or more comorbidities, which was significantly more than survivors without ISF ($p = .022$). The most prevalent comorbidities were back pain (50%), depression (10%), asthma (8%), and thyroid disease (6%). The ISF group contained more survivors with stage II disease or higher ($p = .026$).

Social functioning

The mean social functioning scores of patients with non-ISF and ISF were 96.8 (SD ± 6.6) and 56.1 (SD ± 17.9), respectively. One-quarter of all survivors indicated that their physical condition or treatment resulted in feeling less interested in social activities with other people and 31% of all survivors said their physical condition or treatment resulted in preferring to spend time alone (Table 2). Survivors scored their experienced social support with an average of 8.2, with a lower score reported by the survivors in the ISF group (8.6 vs 7.4, $p < .001$).

Relationships, sexual functioning, and body image

Of all survivors, 73% were in a relationship and there was no statistically significant difference between the non-ISF group and the ISF group (Table 3). Insecurity about their health caused problems in the relationships of 34% of survivors with ISF, compared with 12% for survivors with non-ISF ($p < .001$). The insecurity about their health caused 73% of the survivors with ISF to postpone marriage or committing to a serious relationship, compared with 24% for survivors with non-ISF ($p = .001$).

TABLE 2 Social functioning and experienced social support.

EORTC-CAT-SF	Total *N* = 175 (100)	Nonimpaired SF *N* = 126 (100)[a]	Impaired SF *N* = 49 (100)	*p*
1. Has your physical condition or medical treatment stopped you from looking forward to seeing your family or friends?				
Agree	20 (11.4)	5 (4.0)	15 (30.1)	<.001
Disagree	155 (88.6)	121 (96)	34 (69.9)	
2. As a result of your physical condition or medical treatment, have you felt you did not know what to say to your family or friends?				
Agree	29 (16.6)	16 (12.7)	13 (26.5)	.027
Disagree	146 (83.4)	110 (87.3)	36 (73.5)	
3. As a result of your physical condition or medical treatment, have you preferred to spend time alone?				
Agree	55 (31.4)	24 (19.0)	31 (63.3)	<.001
Disagree	120 (68.6)	102 (81.0)	18 (36.7)	
4. As a result of your physical condition or medical treatment, have you been less able to see your family or friends?				
Agree	33 (18.9)	13 (10.3)	20 (40.1)	<.001
Disagree	142 (81.1)	113 (89.7)	29 (59.9)	
5. As a result of your physical condition or medical treatment, have you felt you were no longer interested in social activities with other people?				
Agree	44 (25.1)	16 (12.7)	28 (57.1)	<.001
Disagree	131 (74.9)	110 (87.3)	21 (42.9)	
6. As a result of your physical condition or medical treatment, have you felt you no longer had a lot in common with your family or friends?				
Agree	32 (18.3)	9 (7.1)	23 (46.9)	<.001
Disagree	143 (81.7)	117 (92.9)	26 (53.1)	
7. As a result of your physical condition or medical treatment, have you spent less time with your family or friends?				
Agree	37 (21.1)	15 (11.9)	22 (44.9)	<.001
Disagree	138 (78.9)	111 (88.1)	27 (55.1)	
8. As a result of your physical condition or medical treatment, have you felt isolated from your family or friends?				
Agree	32 (18.3)	12 (9.5)	20 (40.8)	<.001
Disagree	143 (81.7)	114 (90.5)	29 (59.2)	
9. As a result of your physical condition or medical treatment, have you found it hard to make contact with people?				
Agree	33 (18.9)	11 (8.7)	22 (44.9)	<.001
Disagree	142 (81.2)	115 (91.3)	27 (55.1)	
10. Has your physical condition or medical treatment interfered with your family life?				
Agree	27 (15.4)	8 (6.3)	19 (38.8)	<.001
Disagree	148 (84.6)	118 (93.7)	30 (61.2)	
11. Has your physical condition or medical treatment interfered with your social activities?				
Agree	53 (30.3)	18 (14.3)	35 (71.4)	<.001
Disagree	122 (69.7)	108 (85.7)	14 (28.6)	
12. Has your physical condition or medical treatment interfered with your relationships with your family or friends?				
Agree	28 (16.0)	6 (4.8)	22 (44.9)	<.001
Disagree	147 (84.0)	120 (95.2)	27 (55.1)	
13. Has your physical condition or medical treatment caused you to argue with your family or friends?				
Agree	14 (8.0)	4 (3.2)	10 (20.4)	.001[b]
Disagree	161 (92.0)	122 (96.8)	39 (79.6)	

TABLE 2 (Continued)

EORTC-CAT-SF	Total N = 175 (100)	Nonimpaired SF N = 126 (100)[a]	Impaired SF N = 49 (100)	p
Experienced social support				
Median (range)	9.00 (0 – 10)	9.00 (0 – 10)	8.00 (0 – 10)	<.001
Mean (±SD)	8.22 (1.963)	8.56 (1.773)	7.37 (2.177)	

Abbreviations: EORTC-CAT-SF, European Organization for Research and Treatment of Cancer Computer Adaptive Test Social Functioning; SF, social functioning.

[a]One missing.

[b]Fisher exact.

Of all survivors, 74% had been sexually active in the past 4 weeks, which was statistically significantly lower for survivors with ISF (76% vs 69%, $p = .032$). Almost one-half of all the survivors (46%) felt physically less attractive as a result of their illness or treatment; in the ISF group, this was more often reported (65%) than in the non-ISF group (39%, $p = .006$).

Workability and financial toxicity

At the time of filling in the questionnaire, 78% of all survivors had a paying job (Table 4). Significantly fewer survivors with ISF had a paying job: 59% compared with 86% in the non-ISF group ($p < .001$).

More survivors with ISF reported financial difficulties caused by their physical condition or medical treatment compared with survivors with non-ISF (21% vs 45%, $p = .002$). Survivors with ISF also reported more often that their physical condition or medical treatment caused them financial difficulties that led to changes in their lifestyle (11% vs 43%, $p < .001$).

Factors associated with impaired social functioning

In a multivariable analysis, multiple factors were associated with ISF (Table 5). Higher experienced social support was associated with non-ISF. For every point that survivors reported lower on a 0 to 10 scale, the odds ratio for ISF was 0.739 (95% CI, 0.570–0.957). Unemployment had an odds ratio of 3.719 (95% CI, 1.261–10.967) for ISF. Having to make lifestyle changes due to financial difficulties caused by one's physical condition or medical treatment was associated with ISF in the multivariable analysis (OR, 3.394; 95% CI, 1.118–10.300).

DISCUSSION

This population-based cross-sectional study among sarcoma survivors showed that 28% of the AYA sarcoma survivors experience ISF. Sociodemographic and tumor characteristics associated with ISF include older age at diagnosis, higher stage of disease, and having comorbidities. Psychosocial factors associated with ISF include lower experienced social support, uncertainty in relationships, negative body image, sexual inactivity, unemployment, and financial difficulties. In a multivariable analysis, unemployment, lower experienced social support, and having to make lifestyle changes because of financial problems caused by one's physical condition or medical treatment were associated with ISF.

More than one-quarter of AYA survivors in this study experienced ISF. This is similar to the results from a recent study on social functioning in AYA cancer survivors, which has shown that 2 years after their initial diagnosis, 32% of AYA cancer survivors report consistently low social functioning.[12] In this study, there was no significant difference in time since diagnosis between the non-ISF group and ISF group. This suggests that a large group of survivors struggle with ISF long after diagnosis and that spontaneous improvement is unlikely to occur. However, a longitudinal study has shown that there is a certain degree of fluctuation over time,[12] which suggests that interventions may be successful.

As a result of their disease or treatment, many survivors with ISF preferred to spend time alone (64%) and were no longer interested in social activities (57%). A study among AYA survivors of childhood leukemia described that late effects, such as comorbidities, limited their participation in peer activities. In the univariate analysis conducted in this study, a significantly higher number of comorbidities was found in survivors with ISF, which might partly explain the frequent social isolation in that group. Additionally, survivors in that study mentioned feeling both more and less mature than their peers, less interested in certain typical adolescent social activities (e.g., drinking alcohol, going to parties), and less interested in superficial social relationships, which could all affect social functioning and participation.[22] Survivors with ISF in this study also indicated they find it difficult to make contact with others. A possible explanation for that could be the long time in social isolation during treatment. A study among survivors of childhood brain tumors described how young people can find it difficult to engage with others again after this period.[23]

The multivariable analysis conducted here showed that lower experienced social support is associated with ISF. A previous study in AYA survivors found an association between experienced social support and social functioning in a univariate analysis.[12] Another study among AYA cancer survivors found the same result in a univariate analysis; however, in a multivariable model they did not find that experiencing social support contributed to explaining social

TABLE 3 Relationships, sexual functioning, and body image.

Relationships	Total N = 176 (100)	Nonimpaired SF N = 126 (100)	Impaired SF N = 49 (100)	p
Are you married, do you live together or are you in a long-distance relationship?				
Yes	127 (72.6)	89 (70.6)	38 (77.6)	.357
No	48 (27.4)	37 (29.4)	11 (22.4)	
Missing	1	1	0	
Partnered	**Total N = 127 (100)**	**Nonimpaired SF N = 89 (100)**	**Impaired SF N = 38 (100)**	**p**
I am open and willing to discuss my cancer with my partner.				
Agree	122 (96.1)	85 (95.5)	37 (97.4)	>.999[a]
Disagree	1 (0.8)	1 (1.1)	0 (0.0)	
Neutral	4 (3.1)	3 (3.4)	1 (2.6)	
My partner is open and willing to discuss my cancer with me.				
Agree	116 (91.3)	82 (92.1)	34 (89.6)	.769[a]
Disagree	6 (4.7)	4 (4.5)	2 (5.3)	
Neutral	5 (3.9)	3 (3.4)	2 (5.3)	
Uncertainty about my health has created problems in my relationship.				
Agree	24 (18.9)	11 (12.4)	13 (34.2)	.001[a]
Disagree	90 (70.9)	72 (80.9)	18 (47.4)	
Neutral	13 (10.2)	6 (6.7)	7 (18.4)	
I worry about my partner leaving me if I were to become ill again.				
Agree	4 (3.2)	2 (2.2)	2 (5.2)	.716[a]
Disagree	118 (92.9)	83 (93.3)	35 (92.2)	
Neutral	5 (3.9)	4 (4.5)	1 (2.6)	
Not partnered	**Total N = 48 (100)**	**Nonimpaired SF N = 37 (100)**	**Impaired SF N = 11 (100)**	**p**
Uncertainties about health/future have made me delay relationship.				
Agree	17 (35.4)	9 (24.3)	8 (72.7)	.016[a]
Disagree	23 (47.9)	21 (56.8)	2 (18.2)	
Neutral	8 (16.7)	7 (18.9)	1 (9.1)	
I wonder how to tell a potential partner that I had cancer.				
Agree	16 (33.3)	12 (32.4)	4 (36.4)	>.999[a]
Disagree	27 (56.3)	21 (56.8)	6 (54.5)	
Neutral	5 (10.4)	4 (10.8)	1 (9.1)	
I worry about not having a partner.				
Agree	21 (43.8)	15 (40.5)	6 (54.5)	.724[a]
Disagree	20 (41.7)	16 (43.2)	4 (36.4)	
Neutral	7 (14.6)	6 (16.2)	1 (9.1)	
Sexual functioning	**Total N = 176 (100)**	**Nonimpaired SF N = 127 (100)**	**Impaired SF N = 49 (100)**	**p**
In the past 4 weeks: Have you had decreased libido?				
Yes	83 (47.2)	41 (32.3)	26 (53.1)	.038
No	67 (38.1)	65 (51.2)	18 (36.7)	
Not applicable	26 (14.8)	21 (16.5)	5 (10.2)	

TABLE 3 (Continued)

Sexual functioning	Total N = 176 (100)	Nonimpaired SF N = 127 (100)	Impaired SF N = 49 (100)	p
During the past 4 weeks: Have you been sexually active?				
Yes	131 (74.4)	97 (76.4)	34 (69.4)	.032
No	22 (12.5)	11 (8.7)	11 (22.4)	
Not applicable	23 (13.1)	19 (15.0)	4 (8.2)	
Body Image	**Total N = 176 (100)**	**Nonimpaired SF N = 127 (100)**	**Impaired SF N = 49 (100)**	***p***
During the past week: Have you felt physically less attractive as a result of your illness or treatment?				
Yes	81 (46.0)	49 (38.6))	32 (65.3)	.006
No	81 (46.0)	67 (52.8)	14 (28.6)	
Not applicable	14 (8.0)	11 (8.7)	3 (6.1)	
During the past week: Have you been dissatisfied with your body?				
Yes	95 (54.0)	63 (49.6)	32 (65.3)	.164
No	69 (39.2)	55 (43.3)	14 (28.6)	
Not applicable	12 (6.8)	9 (7.1)	3 (6.1)	
During the past 4 weeks: Have you felt less masculine/feminine as a result of your disease or treatment?				
Yes	58 (33.0)	33 (26.0)	25 (51.0)	.004
No	105 (59.7)	85 (66.9)	20 (40.8)	
Not applicable	13 (7.4)	9 (7.1)	4 (8.2)	

Abbreviation: SF, social functioning.

[a]Fisher exact.

functioning.[24] They argued that experiencing social support might not mitigate interference from fatigue and distress in involvement in social activities.

Interestingly, whether or not survivors were in a relationship did not differ significantly between survivors with ISF and non-ISF. Previous research has shown the benefit of emotional support for AYA survivors in a relationship.[25] Survivors with ISF reported much more often that the uncertainty about their disease and their future caused problems in their current relationship or caused them to delay possible future relationships.

Feeling less attractive was significantly more often reported by survivors with ISF. Previous studies have shown that body image and body perception are frequently affected in AYA cancer survivors.[9,26] This can have far-reaching consequences on psychosocial domains such as self-esteem, identity, and sexuality.[27,28] The results from this study are in line with these previous findings and additionally suggest an association between social functioning and feeling less attractive.

Unemployment was strongly associated with ISF in the multivariable analysis. Based on the results from this study, returning to work in a fitting manner could potentially benefit survivors' social functioning greatly. This might also help reducing financial difficulties that were associated with ISF. Having to make lifestyle changes because of financial problems caused by one's physical condition or medical treatment was also strongly associated with ISF in the multivariable analysis. This supports the hypothesis that financial strain leads to less participation in social activities. As a result, survivors with more financial difficulties and distress could be more at risk for social isolation and ISF.[29]

Limitations of this study include a possible selection bias because it is unknown whether survivors did not participate from an absence of symptoms or a high burden of symptoms. A nonresponder analysis of the SURVSARC study has been published elsewhere and showed no significant difference in disease stage between responders and nonresponders.[21] The cross-sectional character of this study is also a limitation. There is no baseline measurement available for these participants and therefore we do not know how they functioned socially before their sarcoma diagnosis. This was partly compensated for by several items from the EORTC Computer Adaptive Test social functioning and the Workability Index Questionnaire that inquire about the change in functioning caused by a cancer diagnosis. An important limitation of this study regards the single items used to measure the various concepts related to social functioning. Although all of the included items originate from validated questionnaires, the individual items were not always validated as standalone items.

Despite these limitations, to our knowledge, this is the first study to investigate the social functioning of AYA sarcoma survivors. Although social functioning has been investigated before in AYAs, this study provides a level of detail regarding social functioning and

TABLE 4 Workability and financial toxicity.

Workability	Total N = 176 (100)	Nonimpaired SF N = 127 (100)	Impaired SF N = 49 (100)	p
At this moment, do you have a paid job?				
Yes	138 (78.4)	109 (85.8)	29 (59.2)	<.001
No	38 (21.6)	18 (14.2)	20 (40.8)	
How well are you capable of doing your job, compared to before you had cancer? (0–10)				
Median (range)	9 (2–10)	9 (3–10)	7 (2–10)	<.001
Mean (±SD)	8.47 (1.7)	8.88 (1.42)	6.97 (1.86)	
Financial toxicity	**Total N = 176 (100)**	**Nonimpaired SF N = 127 (100)**	**Impaired SF N = 49 (100)**	**p**
Has your physical condition or medical treatment caused you financial difficulties?				
Yes	48 (27.7)	26 (21.0)	22 (44.9)	.002
No	125 (72.3)	98 (79.0)	27 (55.1)	
Missing	3	3	0	
As a result of your physical condition or medical treatment, have you had any extra expenses (e.g., for medication, transport, aids)?				
Yes	79 (45.7)	53 (42.7)	26 (53.1)	.220
No	94 (54.3)	71 (57.3)	23 (46.9)	
Missing	3	3	0	
As a result of your physical condition or medical treatment, have you had extra expenses you had difficulties paying?				
Yes	42 (24.3)	25 (20.2)	17 (34.7)	.045
No	131 (75.7)	99 (79.8)	32 (65.3)	
Missing	3	3	0	
As a result of your physical condition or medical treatment, have you lacked money to buy basic things like food or clothes?				
Yes	14 (8.1)	6 (4.8)	8 (16.3)	.025[a]
No	159 (91.9)	118 (95.2)	41 (83.7)	
Missing	3	3	0	
As a result of your physical condition or medical treatment, have you found yourself in debt?				
Yes	12 (6.9)	7 (5.6)	5 (10.2)	.324[a]
No	161 (93.1)	117 (94.4)	44 (89.8)	
Missing	3	3	0	
Has your physical condition or medical treatment caused you financial difficulties leading to changes in your lifestyle?				
Yes	35 (20.2)	14 (11.3)	21 (42.9)	<.001
No	138 (79.8)	110 (88.7)	28 (57.1)	
Missing	3	3	0	
As a result of your physical condition or medical treatment, have you had less money to spend on yourself (e.g., for buying yourself something that you would like to have but do not necessarily need)?				
Yes	37 (21.4)	19 (15.3)	18 (36.8)	.002
No	136 (78.6)	105 (84.7)	31 (63.3)	
Missing	3	3	0	
As a result of your physical condition or medical treatment, have you had difficulties paying any of your regular expenses (e.g., rent, insurance, phone)?				
Yes	18 (10.4)	8 (6.5)	10 (20.4)	.007
No	155 (89.6)	116 (93.5)	39 (79.6)	
Missing	3	3	0	
As a result of your physical condition or medical treatment, have you had to borrow money or sell personal belongings?				

TABLE 4 (Continued)

Financial toxicity	Total N = 176 (100)	Nonimpaired SF N = 127 (100)	Impaired SF N = 49 (100)	p
Yes	14 (8.1)	7 (5.6)	7 (14.3)	.070[a]
No	159 (91.9)	117 (94.4)	42 (85.7)	
Missing	3	3	0	

Abbreviation: SF, social functioning.

[a]Fisher exact.

TABLE 5 Factors associated with impaired social functioning.

Risk factor	OR	95% CI	p
Older at diagnosis[a]	1.067	.992–1.148	.080
Better experienced social support[b]	.739	.570–.957	.022
Comorbidities ≥1	1.793	.713–4.510	.215
Stage II or higher	2.284	.901–5.793	.082
Feeling less masculine/feminine	1.852	.727–4.717	.196
Not sexually active	2.288	.695–7.536	.174
Unemployed	3.719	1.261–10.967	.017
Lifestyle changes because of financial difficulties caused by physical condition/medical treatment	3.394	1.118–10.300	.031

Note: Multinominal logistic regression analysis among adolescent and young adult sarcoma survivors for the odds of (1) having impaired social functioning vs (0) not having impaired social functioning.

[a]The OR for every year that a survivor is older.

[b]The OR for reporting a point higher on the 10-point scale of experienced social support.

aspects related to social functioning that is rarely seen. Social functioning in AYAs is often researched in common cancers or in cancer in general, yet this provides insights for AYAs with rare cancers that comes with additional obstacles. Another strength of this study is that it was possible to determine if participants had non-ISF or ISF based on age- and sex-matched norm data. This study is population-based, and it includes relatively many survivors of a very rare disease in an age group in which cancer is already uncommon.

These results emphasize the importance of the implementation of age-adjusted psychosocial care in the routine follow-up care of AYA patients. The benefits of acknowledging social issues are potentially lifelong in these young survivors. However, it is a difficult to integrate the detection of these HRQOL issues into the current health care system. It could be helpful to systematically incorporate Patient Reported Outcome Measures into clinical care. In terms of interventions, the AYA care network in the Netherlands is a good example of patient-centered and nurse-led care, focusing on HRQOL in AYA cancer survivors. As the pressure on health care continues to increase, mobile applications that connect cancer survivors with each other and offer tools and tips to potentially improve their HRQOL are highly desirable. For future research, it would be interesting to explore the insecurities surrounding (future) relationships among AYA cancer survivors in a qualitative setting. Also, a longitudinal study design where a baseline measurement is done at the time of diagnosis would be another good method to make claims about the effect of the cancer diagnosis on an individual survivor.

CONCLUSION

This population-based study shows that more than one-quarter of AYA sarcoma survivors experience ISF long after diagnosis. Unemployment, low experienced social support, and having to make lifestyle changes because of financial problems caused by one's physical condition or medical treatment were strongly associated with ISF. These results emphasize the importance of follow-up care for patients with sarcoma that is not only disease-oriented but also focuses on age as well as psychological and social domains.

AUTHOR CONTRIBUTIONS

Cas Drabbe: Data curation, formal analysis, methodology, visualization, and writing – original draft. **Elena S. Coenraadts:** Data curation, formal analysis, methodology, visualization, and writing – original draft. **Winan J. van Houdt:** Writing – review and editing. **Michiel A. J. van de Sande:** Writing – review and editing. **Johannes J. Bonenkamp:** Writing – review and editing. **Jacco J. de Haan:** Writing – review and editing. **Johanna W. M. Nin:** Writing – review and editing. **Cornelis Verhoef:** Writing – review and editing. **Winette T. A. van der Graaf:**

Conceptualization, funding acquisition, investigation, methodology, supervision, and writing – review and editing. **Olga Husson:** Conceptualization, funding acquisition, investigation, methodology, supervision, and writing – review and editing.

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CONFLICTS OF INTEREST STATEMENT

The authors declare no conflicts of interest.

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SUPPORTING INFORMATION

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