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Psychosocial aspects of adolescents and young adults with cancer

Edited by
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and Martha Grootenhuis

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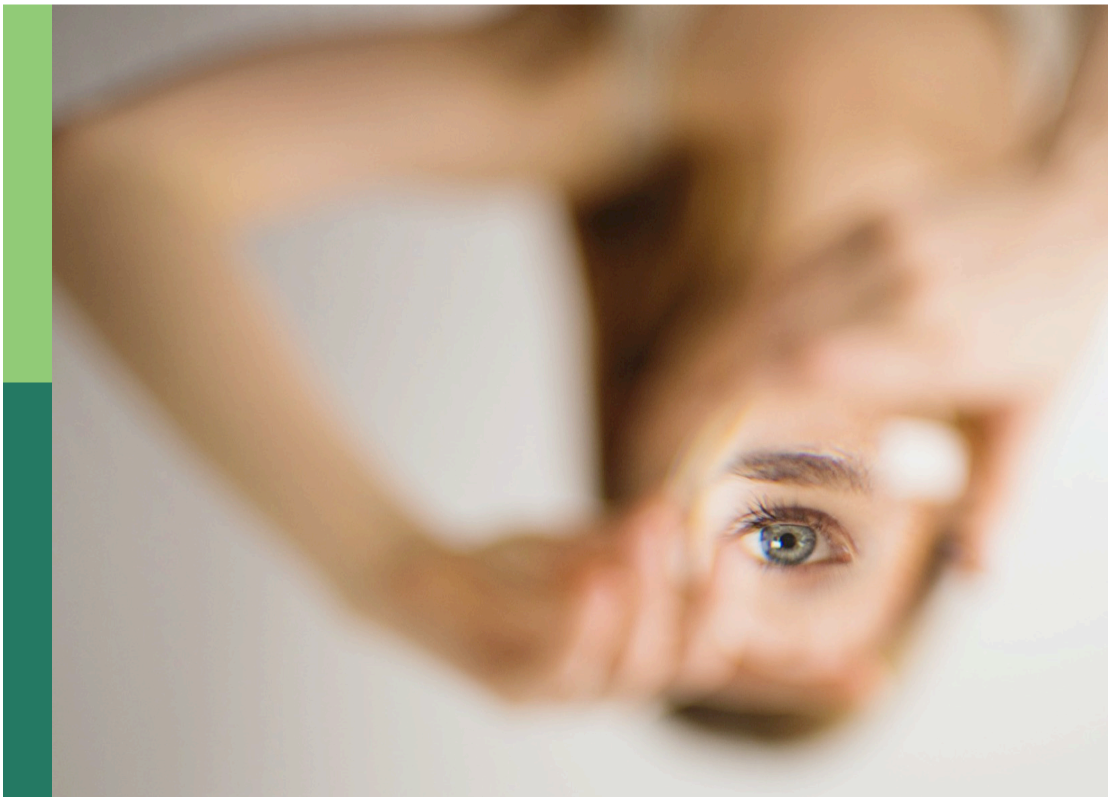

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Psychosocial aspects of adolescents and young adults with cancer

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Editorial: Psychosocial aspects of adolescents and young adults with cancer

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Editorial on the Research Topic
Psychosocial aspects of adolescents and young adults with cancer

Broadly speaking, AYA oncology encompasses clinical care and/or research of individuals diagnosed with cancer between the ages of 15–39 years old. Although varying definitions of the age range are applied, there is no disputing that AYA patients diagnosed or living with cancer (abbreviated as "AYAs" hereafter) experience psychosocial challenges. The AYA period marks a time of tremendous physical-, neurobehavioral-, and social change. Although a continual quest to improve the survival rates of AYA cancers is unquestionably necessary, the unique psychosocial challenges of AYA cancer should also be addressed. This Special Series of *Frontiers in Psychology* highlights research on the unique psychosocial issues faced by AYAs with cancer and their caregivers. Five major topics addressed in this special issue include: (1) the impact of cancer and treatment on AYAs' cognitive, mental, and social well-being, (2) impact of cancer health disparities on psychosocial outcomes among vulnerable groups, (3) informational needs among AYAs, (4) ways to better support family/informal caregivers of AYAs in their caregiving role, and (5) the impact of COVID on the psychosocial outcomes of AYAs.

Focusing on the quality of life aspect of AYA cancer, Tremolada et al. compared cognitive functioning and psychological distress in 205 AYAs and 205 controls. While AYA survivors had lower educational achievement than controls, they reported fewer cognitive problems, and similar psycho-social well-being. Two studies reported on short-term changes in quality of life during the first year of cancer treatment in AYA patients with sarcoma (Day et al.) and young women with breast cancer (Al-Kaylani et al.). Both studies found evidence of short-term improvements in quality of life. Al-Kaylani et al. attribute these early improvements to a sense of relief that may initially outweigh the negative late effects of treatment. These findings underscore the potential importance of identifying AYAs who are at risk of developing psychosocial distress in a clinical setting. The study by Patterson et al. focused on the potential clinical utility and sensitivity of the AYA Psycho-Oncology Screening Tool (AYA-POST). The authors conclude that the AYA-POST can be useful in identifying unique concerns of AYA cancer patients (Patterson et al.). Taken together, these findings suggest that while poor perception of health might be detected in certain subgroups of AYAs during cancer diagnosis and the active treatment phase, their quality of life generally improved during the early post-treatment phase. Since it is still questionable as to whether deficits in quality of life and functioning will emerge during the later survivorship phase, future studies should also evaluate the changes in functional capacity among AYA survivors, such as their work productivity, employment status, and financial security.

Two studies in this series addressed disparity in care (Schwartz-Attias et al.; Kivlighan et al.). The study by Kivlighan et al. focuses on sex-based differences in access to care, where female AYAs were 2.5 times more likely to be referred to behavioral oncology services than male AYAs. The authors call for increased recognition of sex-based biases in referring patients to the appropriate long-term care.

Schwartz-Attias et al. highlight a particularly relevant psychosocial aspect of AYA cancer: health-information found on the internet. The authors' qualitative study revealed that younger AYA cancer patients (15–18 years old) faced challenges in assessing the credibility of the information they received from the internet, and preferred an open discussion with medical professionals. These studies are timely reminders that it is important to consider demographic and sociocultural disparities in AYA psychosocial oncology, especially in developing age-appropriate, gender-specific, and culturally sensitive interventions to address the informational needs and psychological challenges faced by AYAs.

Studies by Melguizo-Garín et al. and Bedoya et al. emphasize the importance of peer/community support in the caregiving journey, and provides important directions in developing family-centered social support interventions for AYAs. Being diagnosed with cancer during a period of significant physical and psychological alterations can create an overwhelming amount of stress to the affected individual, as well as their family/informal caregivers (a role often assumed by a member of their family, companion, or a close friend). Melguizo-Garín et al. demonstrated the positive impact of receiving social support on reducing stress levels in families of AYAs. In line with the notion of "paying it forward," the same study also found that the act of extending peer support to other families in need, helped to enhance life and family satisfaction among caregivers. When discussions on end-of-life care is inevitable for AYAs who have poor prognosis, Bedoya et al. highlighted the value of having a systematic- and research-informed advance care planning in enhancing open communication among AYAs, family members and friends.

Lastly, the COVID-19 pandemic has created unprecedented challenges in cancer care over the past 3 years. Anxieties related to infection susceptibility, missed appointments due to lockdowns, and uncertainties regarding vaccinations are particularly relevant to AYAs and their caregivers. Nearly half of the AYAs in the study cohort of Hou et al. described that their mental health status as worse now than before the pandemic, while Guido et al. reported that parents with symptoms of post-traumatic stress related to their child's diagnosis appeared to be even more vulnerable to stress symptoms perceived during the pandemic lockdown. Fortunately, the availability of vaccinations and effective viral medications have led to the gradual return to normalcy in most countries/regions. Despite the resolution of this health emergency, there is still an urging need to continuously monitor the traumatic, lasting impact of the pandemic on AYAs and their caregivers.

To conclude, this Special Series presents current evidence and highlights the need for novel research directions on the field of AYA psychosocial oncology. The impact of these psychosocial, cognitive, and behavioral outcomes on AYAs' vocational and/or occupational achievements and functional independence requires further investigation. Future work should aim to address the unique psychosocial needs of AYAs that belong to underserved communities, such as ethnic minorities, LGBTQIA+ AYAs, and individuals from rural communities.

Author contributions

YC and EP: preparation of manuscript draft. All authors review and editing. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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BRIEF RESEARCH REPORT
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Psychological Impact of COVID-19 on Parents of Pediatric Cancer Patients

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The changes and general alarm of the current COVID-19 pandemic have amplified the sense of precariousness and vulnerability for family members who, in addition to the emotional trauma of the cancer diagnosis, add the distress and fear of the risks associated with infection. The primary objectives of the present study were to investigate the psychological impact of the COVID-19 pandemic on the parents of pediatric cancer patients, and the level of stress, anxiety, and the child's quality of life perceived by the parents during the COVID-19 epidemic. The parents of 45 consecutive children with solid and hematological tumors were enrolled. Four questionnaires (Impact of Event Scale-Revised – IES-R; Perceived Stress Scale – PSS; Spielberger State – Trait Anxiety Inventory – STAI-Y; Pediatric Quality of Life Inventory – PedsQL) were administered to the parents at the beginning of the pandemic lockdown. A 75% of parents exhibited remarkable levels of anxiety, with 60 subjects in state scale and 45 subjects in trait scale having scores that reached and exceeded the STAI-Y cut off. The bivariate matrix of correlation found a significant positive correlation between the IES-R and PSS scores ($r = 0.55$, $P < 0.001$). There was a positive correlation between the PSS and PedsQL (emotional needs) scale ($P < 0.001$) and a negative correlation between IES-R and STAI-Y ($P < 0.001$). The results confirm that parents of pediatric cancer patients have a high psychological risk for post-traumatic symptoms, high stress levels, and the presence of clinically significant levels of anxiety.

Keywords: psycho-oncology, pediatric oncology, COVID-19, parent perception, cancer, children, stress, quality of life

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INTRODUCTION

The COVID-19 pandemic has affected several aspects of lives all around the globe, and the unprecedented health crisis has put a strain on health services. The literature shows that lockdown measures can affect mental health with several psychological consequences: anxiety, stress, depression, frustration, irritability, insomnia, post-traumatic stress symptoms, and anger (Brooks et al., 2020; Di Giuseppe et al., 2020; Franceschini et al., 2020; Osimo et al., 2021). In Italy and Spain children show increasing screen time, less physical activity, and more sleep; many parents reported changes in their children's behavior and emotional state (Ferrari et al., 2020; Orgilés et al., 2020) and higher levels of parental burnout were reported, especially parents of children with a mental or physical disorder (Fontanesi et al., 2020).

Cancer patients were particularly affected, due to their vulnerability, immunosuppression, or need for cancer treatment (Tsamakis et al., 2020) resulting in a high psychological impact (Jones et al., 2020). Among oncology patients, infants and children are at higher risk for medical or psychological complications (Bitsko et al., 2016; Brinkman et al., 2018). The psycho-evolutionary implications of antineoplastic treatments are well known (Moore, 2005; Oppenheim, 2007; Miller et al., 2009; Brand et al., 2017; Stavinoha et al., 2018), in fact pediatric cancer patient is exposed to continuous events over time that can fall within the field of traumatic stress (Bertolotti et al., 2017). Clinical experience in pediatric oncology shows that trauma can cause psychopathological conditions in survivors (Stuber et al., 2010; Clerici et al., 2014) and also described in the literature (Axia, 2004; Guarino, 2006; Bertolotti and Massaglia, 2011; Hildenbrand et al., 2011), framing pediatric cancer as a stressful and traumatic life cycle event (Patenaude and Kupst, 2005; Phipps et al., 2005; Currier et al., 2009).

The COVID-19 epidemic can represent a further stressful event that is part of a vulnerability framework of the pediatric cancer patient, constituting an additional psycho-pathological risk factor. The researchers on severe psychological trauma (Liotti, 2004; Fosha et al., 2009; Nijenhuis and van der Hart, 2011) and child trauma expert (Lanius et al., 2010, 2012) describe "complex trauma" (Van Der Kolk, 1996; Cook et al., 2005) such as experience of multiple, chronic and prolonged traumatic events (Van der Kolk, 2005).

The risk of COVID-19 infection, and the unpredictability of relative potential emergencies, could exacerbate the emotional burden on patients and family members during oncological disease and treatment. The changes and general alarm of the current pandemic have amplified the sense of precariousness and vulnerability for family members who, in addition to the emotional trauma of the cancer diagnosis, add the distress and fear of the risks associated with infection. The parents fear the consequences of infection on their child's already fragile state of health as well as potential treatment interruptions or delays.

In addition to the standard complex oncological clinical pathway, they require additional measures of self-protection, social distancing (André et al., 2020), prolonged isolation, and new daily habits (Clerici et al., 2020). Also, hospital rules have become more restrictive, requiring the suspension of some services and limitations to family visitation (Leung et al., 2020). These factors significantly affect the patients and their family's quality of life both during hospitalization and afterward upon discharge.

The primary objectives of the present study was to investigate the psychological impact of the COVID-19 pandemic on the parents of pediatric cancer patients, and to investigate the level of stress, anxiety, and the child's quality of life perceived by the parents during the COVID-19 epidemic. Subsidiary objective of the study was to explore correlations between the results obtained and the variables investigated.

MATERIALS AND METHODS

Our study is a single center prospective observational study; duration 9 months. Parents of pediatric cancer patients were enrolled during the 3 months, June–August 2020. Subsequently, the sample was distributed in two groups: parents of patients in treatment (GT) and parents of patients in off-therapy (GOT). Data from the literature report that the level of anxiety and distress of parents, very hight after the diagnosis of their child, can be reduced already during the first 3 months by up to 66% (Harper et al., 2013; Scarponi et al., 2017). Considering the hypothesis of mild correlation ($r = 0.3$) between Impact of Event Scale-Revised and Perceived Stress Scale, an alpha error = 0.05 (two tailed, probability for rejecting the null hypothesis, type I error rate), and a beta error = 0.20 (probability of failing to reject the null hypothesis under the alternative hypothesis, type II error rate) the calculated sample size was 85 cases (Hulley et al., 2013). We concluded the recruitment of the subjects before the expected number of parents was obtained because the recruitment period had ended. Nevertheless, the results obtained confirm the hypothesis of the study.

Participants

The parents of 45 consecutive children with solid and hematological tumors treated in the Pediatric Oncology, Pediatric Neurosurgery, and Radiotherapy Units of Fondazione Policlinico Universitario A. Gemelli IRCCS in Rome were enrolled in the study. Criteria for selecting the subjects were: (1) parent of a patient with a cancer diagnosis; (2) parent of a patient who was in treatment or had completed their treatment regimen; and (3) parent of patients $\leq$25 years of age. The patients $\leq$25 years of age recruited in the study are those who belong to the Unit as suffering from pediatric cancer in treatment or follow-up. Parents with psychiatric or cognitive disorders or intellectual disability were excluded from the study. The parents recruited in the study were screened at the Psychology Service. Parents who were diagnosed with psychiatric disorder were excluded from the study.

This study was performed in accordance with the Helsinki declaration and approved by the Institutional Review Board. Written informed consent was obtained from all participants.

Measures

Impact of Event Scale-Revised (IES-R)

Impact of Event Scale-Revised (IES-R) is a 22-item, self-report measure (for DSM-IV) that assesses subjective distress caused by traumatic events (Weiss and Marmar, 1997; Weiss et al., 2007). The IES-R measures distress, with three subscales assessing Avoidance, Intrusion, and Hyperarousal. In addition to the three subscale scores, IES-R also gives an overall score of events impact (IES-R total, equal to the sum of the three subscale scores). The cut-off of 33 was adopted to indicate a high risk of PTSD symptomatology, in line with the literature. The

TABLE 1 | Demographic and clinical characteristics.

Parents (n)	80
Patients (n)	45
Age at diagnosis (years) mean range	7.96 ± 5.62 2–21 years
Age at study (years) mean range	13.31 ± 6.86 3–25 years
Gender	
Female	18
Male	27
Cancer diagnosis	
Leukemia	9
Lymphoma	4
Solid tumors	32
Treatment status	
In-treatment	18
Off-therapy	27
Parents (n)	80
Age at study (years) mean range	47.34 ± 6.57 30–50 years
Relationship to patient	
Mother	44
Father	36
Level of schooling	
Secondary school	29
High school and bachelor degree	36
Higher education	15
Employment status	
Housewife	19
Teacher	5
Employee	14
Nurse	5
Free lance	10
Worker	11
Military employee	5
Artisan	9
Unemployed	2
Numbers of children	
Only child	9
More than one child	36

Italian translation of the IES – R showed satisfactory internal consistency in studies on different at-risk populations (Intrusion, α = 0.78; Avoidance, α = 0.72; Hyperarousal, α = 0.83) (Craparo et al., 2013; Forte et al., 2020). The IES-R is very helpful in measuring the effect of distress, and traumas in oncology (Nakajima-Yamaguchi et al., 2016).

Perceived Stress Scale (PSS)

Perceived Stress Scale (PSS) is a psychological instrument for measuring the perception of stress. The questions ask about feelings and thoughts during the previous few months (Mondo et al., 2019; Cusinato et al., 2020). The PSS-10 is a self-report instrument consisting of 10 items. Each of the items on the PSS-10 are rated on a 5-point Likert scale, ranging from 0 (never) to 4 (very often). The PSS-10 consisted of 6 positively (items 1, 2, 3, 6, 9, and 10: Positive factor) and 4 negatively (items 4, 5, 7, and 8: Negative factor) worded items. Total scores range from 0 to 40, with higher scores indicating higher levels of perceived stress. Scores ranging from 0 to 13 would be considered low stress. Scores ranging from 14 to 26 would be considered moderate stress. Scores ranging from 27 to 40 would be considered high perceived stress. It was frequently used during the pandemic in Italy and other countries (Limcaoco et al., 2020; Rossi et al., 2020). Internal consistency estimates using Cronbach's alpha range from 0.67 to 0.91.

Spielberger State – Trait Anxiety Inventory (STAI-Y)

Spielberger State – Trait Anxiety Inventory (STAI-Y) is a 40-item, self-completed questionnaire that aims to separately assess state anxiety (STAI-Y1, a temporary state influenced by the current situation) and trait anxiety (STAI-Y2, a general propensity to be anxious) with 20 items each (Pedrabissi and Santinello, 1989; Cafisio and Tralongo, 2004). Scores over 40 on both the state and trait scales were adopted; this value corresponded to the point at which false positive and negative results were minimal (Barnett and Parker, 1986; Hart and McMahon, 2006). The internal consistency reliability ranges from 0.91 to 0.95 for the scale of state and from 0.85 to 0.90 for the scale of trait.

Pediatric Quality of Life Inventory^{TM} (PedsQL 4)

Pediatric Quality of Life Inventory^{TM} (PedsQL) 4.0 Generic Core Scales is a parent proxy-report including Physical, Emotional, Social, and School Functioning Scales. It assesses parents' perception of their child's Health-Related Quality of Life. Higher scores represent better quality of life. It has been used frequently and is well validated within pediatric oncology populations. The PedsQL has demonstrated good psychometric properties across studies including Cronbach's alphas that met or exceeded 0.70 and good construct validity in pediatric cancer samples (Varni et al., 1999; Racine et al., 2018). The scale has good internal consistency reliability for the total scale score (alpha = 0.90 on parent report).

Procedure

The questionnaires were administered to the parents at the beginning of the pandemic lockdown. The researchers explained the purpose of the study to the parents.

Their written informed consent to participate in the study was obtained, and they were reassured about the confidentiality of the information they provided. Most parents preferred to be interviewed rather than to complete the questionnaires on their own. For each patient included in the study, the questionnaires were individually administered to the parents. The parents were informed that the IES-R scale referred to their child's cancer diagnosis, while the other questionnaires referred to the current phase of the pandemic.

Statistical Analysis

Correlations between the scores of the 4 scales were analyzed. Subsequently, the correlation between the questionnaires (IES-R, PSS, STAI-Y, PedsQL) and the variable "months," the time

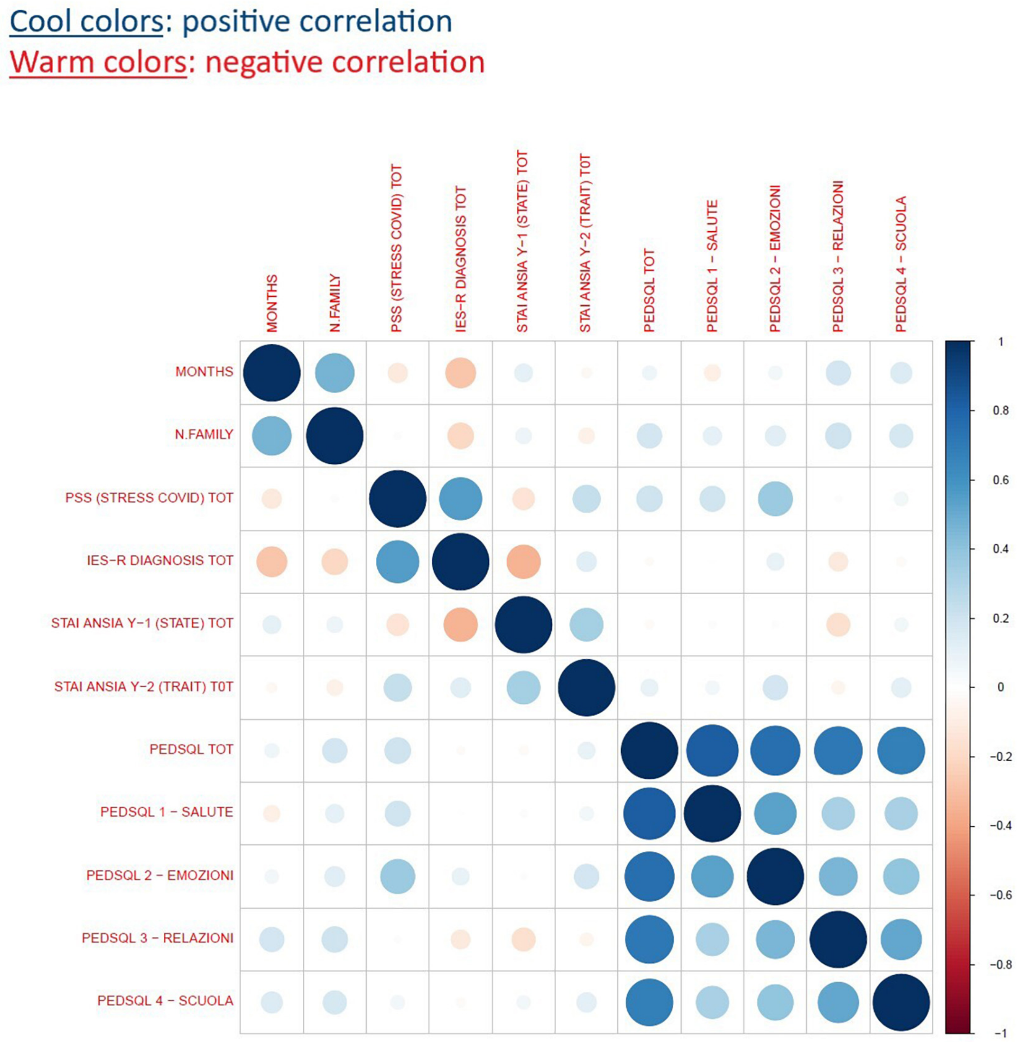

FIGURE 1 | Cross correlation matrix.

between oncological diagnosis to starting the study, was determined. Comparisons between groups of parents were made using the Mann-Whitney *U* test for non-parametric samples. Statistical analysis was performed using R 4.0.3 version.

RESULTS

The study included the parents of 45 patients (32 with solid tumors and 13 with malignant hematological diseases). They were divided into 2 groups, those who had completed their treatment regimen (off-therapy group, $n = 27$) and those who were still receiving treatment (in-treatment group, $n = 18$). There were 18 females and 27 males.

One father had died, 1 father had psychiatric disorders, and 5 parents (1 mother and 4 fathers) did not fill in the questionnaires. Therefore, a total of 80 parents (44 mothers and 36 fathers) were included in the study. All parents agreed to participate and provided written informed consent. Demographic characteristics of the participants are summarized in **Table 1**.

A total of 87.5% of the parents had a moderate ($n = 20$) or high ($n = 50$) risk for traumatic disorder (IES-R, $x = 41.68 \pm 16.72$), and 83.7% had a moderate ($n = 54$) or high ($n = 13$) presence of stress symptoms (PSS, mean = 19.25 ± 5.33). In our sample 75% of parents exhibited remarkable levels of anxiety, with 60 subjects in state scale and 45 subjects in trait scale having scores that reached and exceeded the STAI-Y cut off. The mean values were Y1 (state), $x = 42.48 \pm 4.32$ and Y2 (trait), $x = 41.15 \pm 4.56$.

The bivariate matrix of correlation (**Figure 1** and **Table 2**) found a strong significant positive correlation between the IES-R and PSS scores ($r = 0.55$, $P < 0.001$). There was a positive correlations between the PSS and PedsQL (emotional needs) scale ($P < 0.001$) and a negative correlation between IES-R and STAI-Y ($P < 0.001$).

It was not possible to make comparisons between the parents of patients diagnosed during and before the pandemic due to

TABLE 2 | Cross correlation matrix coefficients (up) and Pearson correlation tests *P*-Values (down) in each item of the table.

Correlation matrix											
	1	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**
(1) MONTHS	1										
(2) N. FAMILY	0.46704 0.00001	1									
(3) PSS (STRESS COVID) TOT	−0.11761 0.30196	0.01631 0.88658	1								
(4) IES-R DIAGNOSIS TOT	−0.27920 0.01270	−0.20633 0.06810	0.55555 <0.00001	1							
(5) STAI ANSIA Y-1 (STATE) TOT	0.10116 0.37499	0.07998 0.48349	−0.14880 0.19060	−0.347998 0.00167	1						
(6) STAI ANSIA Y-2 (TRAIT) T0T	−0.03247 0.77632	−0.07877 0.49015	0.23348 0.03837	0.122036 0.28399	0.33765 0.00234	1					
(7) PEDSQL TOT	0.06279 0.58248	0.18741 0.09816	0.20701 0.06718	−0.020476 0.85785	−0.02569 0.82221	0.09087 0.42577	1				
(8) PEDSQL 1 – SALUTE	−0.08289 0.46767	0.10616 0.35176	0.19694 0.08192	−0.006373 0.95555	0.01834 0.87255	0.05999 0.59942	0.82701 <0.00001	1			
(9) PEDSQL 2 – EMOZIONI	0.05959 0.60192	0.12658 0.26631	0.36331 0.00099	0.092094 0.41954	0.01375 0.90424	0.18840 0.09636	0.75521 <0.00001	0.539688 <0.00001	1		
(10) PEDSQL 3 – RELAZIONI	0.18538 0.10191	0.20458 0.07052	0.01428 0.90060	−0.112361 0.32418	−0.16864 0.13737	−0.05471 0.63205	0.71189 <0.00001	0.32832 0.00314	0.45039 0.00003	1	
(11) PEDSQL 4 – SCUOLA	0.14055 0.21664	0.17065 0.13266	0.05980 0.60061	−0.028581 0.80256	0.05759 0.61413	0.11877 0.29717	0.68242 <0.00001	0.327831 <0.00001	0.39479 0.00032	0.51120 <0.00001	1

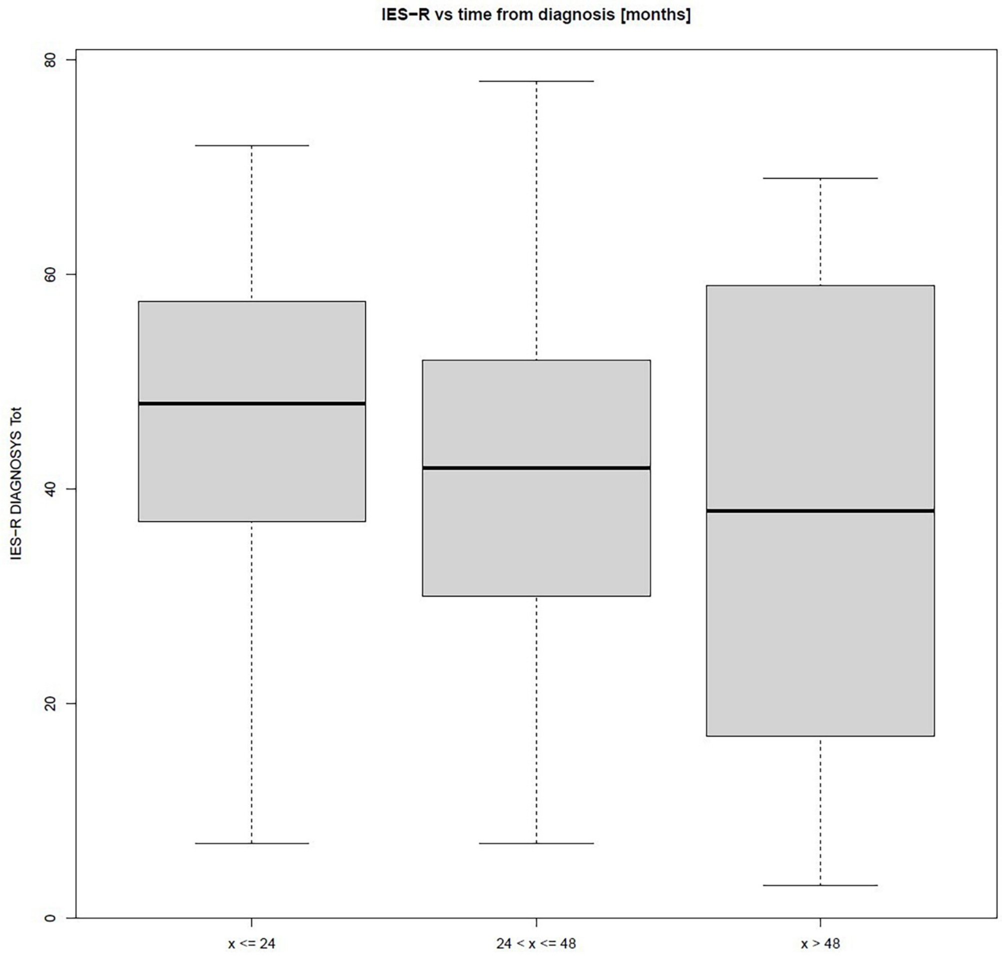

FIGURE 2 | Time from diagnosis and Impact of Event Scale-Revised (IES-R) results.

the inhomogeneity between the groups. However, the 8 parents interviewed whose children were diagnosed during the pandemic had an average IES-R of 50.28. Since there was a weak correlation ($P > 0.05$) between the time from diagnosis to completing the IES-R, the sample was further divided according to the time from diagnosis, <24, 24 – 48, and >48 months. In **Figure 2** the trend line shows a decrease over time, but the difference between the groups is not significant, and in the third group ($x > 48$ months) there is also a large dispersion of scores. Comparisons between the groups were also made separating parents into those with and without high trait anxiety (divided according to the STAI-Y2 cut-off). Even this group of patients did not have significant differences in any of the test variables.

Separating the groups into those off and on therapy found that this variable had a significant impact on the outcome of IES-R ($P < 0.001$; off-therapy, $x = 36.60 \pm 4.84$; on-therapy, $x = 49.55 \pm 16.23$) and PSS ($P < 0.001$; off-therapy, $x = 18.10 \pm 4.84$; on-therapy, $x = 21.03 \pm 5.64$). Subsequent comparisons between groups found a significant difference between the scores of mothers and fathers only on the PSS ($P < 0.001$; mothers, $x = 20.89 \pm 4.90$; fathers, $x = 17.20 \pm 5.21$).

DISCUSSION

The diagnosis of cancer in the pediatric age group is widely described as a traumatic event for the parents, and may result in experiences, emotions, and even symptoms of psychopathological conditions such as post-traumatic stress disorder (Santacroce, 2002; Landolt et al., 2003; van Warmerdam et al., 2019), studies offer discordant results (Norberg and Boman, 2013; Ringnér et al., 2015). Generally, parents' high levels of anxiety and distress following their child's diagnosis (Patiño-Fernández et al., 2008; Vrijmoet-Wiersma et al., 2008) decrease over time with a decline already present 3 months after diagnosis (Harper et al., 2013; Scarponi et al., 2017).

The principal objective of the present study was to explore the psychological impact on parents of children with cancer during the health emergency caused by the COVID-19 pandemic. We were interested in determining if, unlike a period without a socio-sanitary emergency, the influence of post-traumatic experiences might exacerbate the challenges or symptoms, such as anxiety or stress, or place parents at a new or additional risk of psychological suffering (Evans et al., 2020). This supposition is supported by evidence showing that COVID-19 has great emotional impact, even on the general population, with or without specific medical conditions (Sani et al., 2020).

Consistent with our hypothesis, our sample parents showed high levels of post-traumatic symptoms related to the oncological diagnosis of their child, even at a time remote from diagnosis (Ringnér et al., 2015; Ribeiro da Silva, 2018). They also had an elevated perception of stress symptoms referable to the pandemic, much higher than the general population's during COVID-19 (Limcaoco et al., 2020; Pedrozo-Pupo et al., 2020). The average level of state anxiety, measured with STAI-Y1, indicated the presence of a considerable number of anxiety symptoms.

The correlation matrix (two-tailed) showed that parents recording a higher traumatic impact level on the IES-R for child cancer diagnosis also perceived higher stress levels caused by the COVID-19 measured with the PSS. It suggests the possibility of identifying populations at risk for experiencing sequelae and consequences on child well-being. Indeed, studies have shown that parents experiencing greater stress find it more difficult to understand their child's needs and respond in a sensitive manner (Scaramella et al., 2008; Spinelli et al., 2020) and that parenting stress might have detrimental effects on children (Giannotti et al., 2021).

The results of this study show that parents who exhibit symptoms of post-traumatic stress related to their child's diagnosis appear to be more vulnerable to stress symptoms perceived during the pandemic lockdown. The parents' symptom states do not appear to be related to the individual characteristics of anxiety traits. In fact, comparing the scores of "anxious" and "non-anxious" parents, there were no significant differences on all questionnaires except for the form of state STAY-Y 1.

Data from the few parents who received the oncological diagnosis of their child during the pandemic show a high level of PSS. Parents who received the diagnosis close to the onset of the COVID-19 pandemic were subjected to this potential acute stress event (Spinelli et al., 2020) and showed an increase in the already high risk (Santacroce, 2002; Norberg and Boman, 2013) of developing post-traumatic symptoms.

Therefore, it seemed worthwhile to investigate the correlation with temporal distance from the time of diagnosis to understand the role of time as a protective factor (Vrijmoet-Wiersma et al., 2008; Lazor et al., 2019). Among this study's participants, the variable "months" from the time of diagnosis did not have a significant impact on parent score. To understand this phenomenon, it is important to consider that the scores of most parents documented a significant presence of post-traumatic symptoms. In accord with the literature, traumatic psychological conditions can have long term consequences (Porges, 2009; Kolacz et al., 2019). The COVID-19 pandemic has rapidly affected the care for children with cancer worldwide (Bouffet et al., 2020; Graetz et al., 2021) and parent perception of assistance (Guidry et al., 2021; Mirlashari et al., 2021). Italian research also documents downstream consequences on the psychosocial functioning of tumor survivors (Fisher et al., 2021); therefore we were interested to collect parents' impressions of their child's quality of life (van Gorp et al., 2021). Through the inclusion of PedsQL parent proxy-report version made it possible to collect important information on the children's activities and behaviors during COVID-19 (physical, scholastic and social activity) as well as the degree of emotional needs or difficulties of children. This last variable showed a significant positive correlation ($P < 0.001$) with the tool on parental stress (PSS), highlighting a strong relationship between the psychological state of child and parent (Kohlsdorf and Costa Junior, 2012; Salvador et al., 2019; Santos et al., 2019; Tillery et al., 2020). Previous reports have found a significant difference between mothers' and fathers' scores on PSS ($P < 0.001$) (Hoekstra-Weebers et al., 2001; Yeh, 2002; Norberg and Boman, 2013; Compas et al., 2015). Our results also show a significant difference between parents of patients "off therapy" and those still "on therapy" in IES-R ($P < 0.001$) and PSS ($P < 0.001$) scores. Studies show that it is important to observe the stress of parents of children with cancer throughout their lives (Ringnér et al., 2015; Ribeiro da Silva, 2018), but in this particular emergency it seems to be very important to do so during treatment, when children are most at risk of infection (Auletta et al., 2020; Bouffet et al., 2020; Evans et al., 2020; Seth, 2020; Seth et al., 2020).

CONCLUSION

Worldwide, data suggests that pediatric cases of COVID-19 are less severe than adults (Evans et al., 2020). However, the possibility that their child might be infected creates worry and fear in parents, especially if the child has a pre-existing condition such as cancer where infection with COVID-19 might aggravate symptoms and pose an additional risk to the child's health. Preliminary results of our longitudinal study, which will continue to investigate parental symptoms and variables over the course of 9 months, show a significant positive correlation between parental scores of traumatic impact of their child's cancer diagnosis (IES-R) and parental stress perception during the COVID-19 outbreak (PSS).

The COVID-19 pandemic has introduced new challenges for the organization of health services and multidisciplinary work (Amicucci et al., 2020). This study highlights the importance of integrating care for the parents with care for the child through continuous monitoring of their psychological state and the need for parent-oriented interventions.

The present study is limited by the absence of a comparison group of parents who have not experienced the pandemic, and there is no control group of parents whose children do not have a cancer diagnosis. We will attempt to increase the reliability of the investigation by making comparisons between subjects with the re-test that will be performed, according to the study's protocol, in the coming months.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Fondazione Policlinico Universitario Agostino Gemelli IRCCS Rome. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

AG, EM, LP, AR, and DC were involved in study planning and led to the preparation of the manuscript. AG, EM, LP, ND, GT, GA, MB, VV, and DC were involved in study conduct. All authors were involved in the reporting and reviewing of the manuscript.

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Adolescents With Cancer Need Trustworthy Information and Prefer to Receive It From a Human Source Rather Than From the Internet: A Qualitative Study

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Background: In pediatric cancer, the legal obligation to provide information is usually toward the parents who are the authorized signatories of the informed consent form. It is now known that aside from providing information to the parents, it is also very important to provide information to the children and adolescents themselves. The question is how the adolescents relate to this. What information do they already possess and what would they like to know? Would they wish to hear the truth in all situations and at what stage? What are their preferred sources of information?

Method: A qualitative study that included in-depth interviews with 19 adolescents with cancer, aged 8.5–18, who were receiving active treatments and had been diagnosed at least 1 month previously. The interviews were guided by 15 open-ended questions.

Findings: The analysis of the interviews indicated that adolescents know quite a lot about the course of their disease and the information they lack is mainly etiological. The participants reported a lack of knowledge concerning sexuality and a sense of discomfort talking about it, leaving them with open questions. They all claimed that it is important to tell the truth: "Even if the truth is difficult, it is important to tell it." The participants reported that information can be scary, so it must be structured and adapted to the age and emotional readiness of the individual. Most of the participants prefer not to use the internet as an information resource due to the profusion of stressful and non-adapted information.

Conclusion: Adolescents with cancer need trustworthy information and prefer to receive it from a human source rather than from the internet. Not telling the truth can lead them to feel fear and loneliness. The medical staff must operate in sensitive and creative ways to provide adolescents with access to information on various subjects, including sexuality, which they are ashamed to talk about, leaving them with a sense of shame and a lack of knowledge in this area.

Keywords: information needs, adolescents with cancer, sexuality, telling the truth of cancer, information resource

INTRODUCTION

In the patient-centered approach, communication skills constitute significant milestones (Zanon et al., 2020). This approach advocates sharing with the patient all details of the diagnosis, the treatments, and their consequences (Ali, 2017). There is a great deal of information on the significance of adapting the manner of communication of the medical staff to the values and preferences of the patient (Kissane et al., 2010; Brown et al., 2016). Many studies conducted in this area found a correlation between effective personally adapted communication and compliance with treatment and better treatment outcomes (Ali, 2017). Legally, when diagnosing pediatric cancer, the responsibility to provide information regarding the diagnosis and treatments falls on the guardian. The most common guardians are the parents, who are the authorized signatories of the informed consent form for patients under 18. Aside from providing information to the parents, it is very important to provide information regarding the diagnosis and planned treatment to the children and adolescents themselves (Bahrami et al., 2017; Stein et al., 2019). The ability of children to understand the diagnosis and the treatment protocol depends on many factors, including their developmental stage. A study conducted among survivors of childhood cancer found that adolescents who had received structured information at the diagnosis stage (adapted information provided by a member of the medical staff not long after the diagnosis) displayed fewer psychological impairments further on in life than those who had not received such information (Raz et al., 2016).

Information for Adolescents – What Do They Want to Know

Communication with adolescents with cancer is extremely important and can affect their adjustment to the diagnosis and treatments, their satisfaction with the treatment, their decision-making process, and further, their life as healthy individuals in the community (Raz et al., 2016; Bahrami et al., 2017; Bibby et al., 2017). Using various research methods, Decker et al. (2004) investigated the information that adolescents wish to receive about their cancer and how they are affected by the information or the lack thereof. In the quantitative part of the study, considerable significance was attributed to the need for information on handling medical procedures, relations with friends and family, returning to school, and completion of treatment. In the qualitative part, additional issues that occupy affected adolescents were uncovered, including types of treatment and side effects, uncertainty regarding the future, as well as social and emotional matters. In light of the need for information on different topics, including psychosocial aspects, the researchers concluded that ongoing informational support should be provided to the affected adolescents and that they should be provided with information regarding their illness, its treatment, and the physical and emotional implications of this type of challenge, from the moment of diagnosis (Decker et al., 2004).

In the two decades that have elapsed since then, the knowledge base regarding the unique needs of adolescents with cancer has not grown considerably. In their extensive literature review, Bibby et al. (2017) found that most publications in this area do not relate specifically to adolescents with cancer. The review, which included research on adolescents and young adults (AYA) aged 15–30, included 45 articles, where only about one-quarter of them referred to adolescents. The same review indicated that adolescents with cancer report a need for age-adjusted information concerning diagnosis, treatments, maintaining fertility, a healthy lifestyle, and recovery. In addition, adolescents reported that receiving information helps them prepare for the future better, while the lack of information is associated with a sense of stress and dissatisfaction with treatment (Bibby et al., 2017). A qualitative study conducted in Iran among 12 adolescents, aged 15–20, with cancer found that all the participants reported that the information they had received directly about their illness was deficient and that most of the information had been provided to their parents. The adolescents had (many) unanswered questions and were unable to find a reliable source for obtaining answers (Bahrami et al., 2017).

Sexuality Among Adolescents

Sexuality constitutes an important aspect in the development of adolescents and in their process of entering the adult world, and it is associated with their well-being and quality of life (Laumann et al., 1999). The literature indicates that the topic of sexuality continues to occupy the thoughts of many adolescents despite their condition (Veneroni et al., 2020).

Nevertheless, the topic is often considered taboo by health care providers, who are uncomfortable to raise and discuss it openly with adolescents (Graugaard et al., 2018; Veneroni et al., 2020). In their reports, adolescents with cancer raise difficulties related to sexuality, such as altered body image, diminished self-value, decreased sexual drive, the concern of the spouse regarding having sex as before the diagnosis, as well as other concerns (Graugaard et al., 2018; Veneroni et al., 2020). It is concerning that adolescents report a lack of opportunity to discuss these topics. A study conducted among 66 adolescents with cancer, aged 16–24, in Italy, found that 39% of the participants reported that health care providers had never spoken to them about these topics, while 17% claimed that these topics had been discussed with them to a limited extent. Eighty percent of the participants thought that these topics should be given more room in the therapeutic discourse (Veneroni et al., 2020).

Telling the Truth From the Beginning

The issue of telling the truth to patients is the topic of many ethical discussions. Davies (2020) deals with whether it is always right, to tell the truth, what to say and how, and what happens if patients 18 years and older do not want to know, or if the information might harm them. Additionally, what happens when the patient is a minor (under 18) and the obligation of providing the information to the patient is entrusted to the guardian (Zanon et al., 2020). According to the directives developed by the International Society of Paediatric Oncology (SIOP), children and adolescents under 18 with end-stage cancer should be given as much information as possible about their condition in accordance with their age and developmental stage, "Depending on age and level of development, the child should also be involved in the decision, with older children especially participating more actively" (Masera et al., 1999, p. 46). The professional directives of the SIOP from 2009 also state that sick children and adolescents under 18 have a basic right to receive adapted information (Kowalczyk et al., 2009). But these directives are nonetheless very general and leave much room for personal and professional interpretation. A study conducted among 203 AYA aged 15–29 found that patients who received an extensive explanation regarding their prognosis reported more trusting relations with the health care providers, mental calmness, and more hope (Mack et al., 2018). A qualitative study held in Sweden among adolescents with cancer, aged seven to 17, found that they want to hear the truth about their condition, including receiving difficult information. All the participants claimed that no matter how tough the information, they want to know it, but it is important for them that during the conversation they are not denied hope (Jalmsell et al., 2016).

Information Resources Among Adolescents With Cancer

Adolescents raise varied and diverse needs for information. The differences in the consumption of information may be related to age, the stage of the illness, type of disease, and resilience (Lea et al., 2018). One of the challenges confronting health care providers of adolescents with cancer is providing as much accurate and personally adapted information as possible. Each adolescent has different information needs in terms of the content and scope of information. For example, some adolescents will need information related to sexual relationships while others will need information related to obtaining a wig. Some adolescents need broad information and some need limited and superficial information. Naturally, some patients, including children and teens, will seek and gather information on their illness from their surroundings and according to their cognitive abilities. When diagnosing cancer in childhood or adolescence, parents themselves may be in a state of trauma and therefore the responsibility of delivering professional and reliable evidence-based information to sick children and adolescents falls to the medical staff (Zanon et al., 2020).

The internet is an important, readily available, and integral source of information in the life of adolescents, however, despite its advantages this information platform also has its shortcomings. A study conducted among 21 adolescents, aged 13–24, with cancer, found that searching for factual information about their cancer diagnosis, prognosis, treatment, tests, and procedures is complex and problematic. Some young people do not use this resource at all, and some express uncertainty and difficulties regarding the validity and reliability of the information and the professional jargon (Lea et al., 2018). Others report various emotional consequences such as fear, concern, and anxiety.

A study by Raz et al. (2016) examined the impact of the type of information on the emotional state of convalescents. Adolescents who reported that the information they received during their illness had been provided by the medical staff in an orderly manner demonstrated better scores for quality of life, psychological pain, and tolerance for psychological pain than convalescents who reported having obtained this information on their own.

The current study is a pioneer study conducted in Israel and its main purpose was to illuminate the existing and missing types of information and the preferred information resources among adolescents with cancer in active stages of treatment, using qualitative research methods. The study focused on adolescents with cancer, aged 8.5–18 years, an age group considered psychologically unique (Raz et al., 2016; Bahrami et al., 2017; Sawyer et al., 2018). Adolescence is characterized by the transition from childhood to adulthood when adolescents are engaged in seeking independence and establishing their personal and unique identity among other things while demonstrating introspective, abstract, and operational reasoning (Nixon, 2014). Therefore, the question is: What information would adolescents like to receive in the different stages of their illness, what are their preferred information sources, and how would they like to receive the information. In the current study, while receiving treatment, adolescents were asked what information they would like to receive from the moment of diagnosis, what are information resources they use, and what issues occupy them. According to the literature, this important topic has yet to be sufficiently investigated. Observing the life of affected adolescents and becoming familiar with their illness-related content worlds can help health care providers maintain evidence-based communication that may provide a better response to the unique information needs of the former regarding diagnosis and treatment and thus, improve their present and future psychosocial outcomes.

The Research Questions

The main questions we sought to explore in this study were:

(1) What do children and adolescents know about their illness, by age?
(2) What would children and adolescents want to know in general or more than they know at present?
(3) What are the information resources they use and that they would recommend for others in their condition?

MATERIALS AND METHODS

Participants

The study included 19 adolescent cancer patients (**Table 1**) treated at a tertiary pediatric medical center in Central Israel. The inclusion criteria were age ≤18 and ≥8 years, diagnosed with cancer, receiving chemotherapy, and speak Hebrew. Accordingly, the exclusion criteria were as follows: older than 18 or younger than 8, with no cancer. All the participants were Jewish, four reported that they keep a secular lifestyle, and the rest were traditional, religious, or

TABLE 1 | Characteristics of the 19 participating adolescents.

		N
Gender	Male	10
	Female	9
Age	8.5–11.5	3
	12–15	5
	15.5–18	11
Diagnosis	ALL	8
	Lymphoma	3
	Ewing sarcoma	2
	Optic glioma	2
	Rhabdomyo sarcoma	1
	AML	1
	Endometrial carcinoma	1
	Aplastic anemia	1
On active treatment	Yes	19
	No	0

ALL, acute lymphoblastic leukemia; AML, acute myeloid leukemia.

ultra-Orthodox. The proportion of secular participants in the current study is low relative to their proportion in the Israeli population at large.

The participants were recruited using a convenience sampling method. The consent process consisted of three parts - The first part included an explanation provided to the parents about the study. The second part included contacting the children whose parents had given their consent to participate in the study. The children received the same explanation as their parents did and upon expressing their consent, the interview phase began. In the interview phase, the researchers first contacted the 23 parents of the adolescents with cancer who had expressed consent for their child to participate in the study. Of those, only 19 children agreed to participate (82% response rate). The research participants were highly satisfied with the interview and said that they feel like ambassadors and are grateful for the opportunity and privilege of helping advance knowledge in the field for the benefit of future patients.

Research Tool and Data Collection

A semi-structured interview following an interview guide was developed according to the main research questions. The interview guide consisted of 15 open-ended questions (**Table 2**) inspired by previous studies (Raz et al., 2016; Bahrami et al., 2017). At the beginning of the interview session, it was clarified to the participants that they could choose whether to answer the questions or not and that they could add information that matters to them even if not asked for. The face-to-face interviews took place in a private room, in the presence of the interviewing nurse and the adolescents, for about 45 min. To avoid a sense of threat or pressure on the children, the interviews were not recorded but the answers of the children were manually transcribed by the interviewers.

Ethical Considerations

The study was approved by the Helsinki Committee at the Rabin Medical Center and all the procedures required for this study were completed.

Data Analysis

The data were analyzed using conventional qualitative content analysis (Hsieh and Shannon, 2005), where codes were defined during the data analysis and derived from the data. The analysis was done inductively, manually, imposing no categories or theoretical perspectives in advance. The analysis began by reading all the transcripts three times, to get a sense of all the data. Then the categorization guidelines of Hsieh and Shannon (2005) were followed. The recommendations of Morse (2015) for determining rigor in qualitative inquiry were maintained.

The first and second authors, who have rich experience in qualitative research, conducted the analysis. The analysis began by creating an initial coding scheme which was done separately by each member of the research team, which included nurses, a psychologist, and a sociologist. After reaching an agreement on the initial coding scheme, the first and second authors continued to the following stage of the analysis. The next step was to organize groups of codes into meaningful categories and subcategories, develop definitions for each category, and identify examples from the data. The team met several times to debrief and discuss the emergent categories and subcategories in-depth.

In the "Results" section, only a full description of the findings is given, as is customary in qualitative methods, and pseudonyms are used, besides the age of the child, for example (Amir, 17) or Amir (17).

RESULTS

The Information That Children and Adolescents Have About the Disease, Treatments, and Side Effects

When asked about what they know about their illness, the participants could answer in their own words relating to their cancer diagnosis, for example, "a problem with cells" (Johnny, 12). Despite the variety in the level of specification/sophistication of the answers, they were able to correctly identify their organ or body system affected by the illness, for example, the blood or pelvis. "I have acute leukemia, a blood cancer. A condition where T cells in the bone marrow remain young and divide quickly. They occupy the bone marrow and it keeps other cells from thriving" (Jonathan, 16). Eight of the participants ($N = 19$) noted the name of the medical diagnosis or used the term "tumor" or "cancer."

The participants knew what treatments they were receiving and what they would be receiving in the future: "I know I have 17 treatments; then surgery and then 11 more treatments" (Noa, 13). They recognized the names of the medications and the reason for taking them, as well as their side effects: "About the treatments, I know it's chemo, that it destroys the healthy cells as well, and that it has side effects: hair loss, nausea, loss of taste" (Rachel, 15).

TABLE 2 | Fifteen questions that guided the study.

Categories	Question No.	Content
Your illness	1	What do you know now about your illness?
	2	Would you like to know more about your illness than you know now?
	3	What kind of information would you like to be given about your illness?
	4	What do you think you should know about your illness?
Your treatments	5	What do you know now about your treatments?
	6	Would you like to know more about your treatments than you know now?
	7	What kind of information would you like to be given about your treatments?
Other illness	8	What do you think a child/adolescent should know about his illness?
	9	What kind of information is usually given to children and adolescents regarding their illness?
	10	What do you think that children and adolescents in the ward know about their illness?
Other illness and treatments	11	In your opinion, how does the information that children and adolescents receive about their illness and treatments, affect them?
Support resources	12	Who mostly helps you?
	13	What most helps you?
Sources of information	14	From whom did you get the information about your illness and treatments? (1) ___________; (2) __________
	15	Where do you search for information about your illness and treatments? (For example internet, parents, nurses, doctors, etc.)

Children and Adolescents Want to Know More About the Cause of Illness and the Treatments

Some of the participants said they need to understand the cause of their illness: "I would like to know why. How did it suddenly appear?" (Amir, 17); "When I arrived, they didn't explain to me about the disease. I wanted to know what was causing it" (Dina, 17). Five participants were interested in what they could do to heal faster, for example, would a change to their nutrition or lifestyle help?

Nine participants said that it is important to share information with children because it helps them cope. They said that at first, they were apprehensive about the information, but ultimately it was reassuring: "At first, it was hard to accept it [the information], but it was good that I heard it." (Ruth, 13). Dan (12) mentioned that "It is very important to know about the disease. It helps cope." He shared that he wanted to know what caused the illness and how it would affect him at school. However, he would not like to know more about the treatments. Unlike him, most of the participants wanted to understand the treatment plan: "I need to know what I'm going through. not just come to the hospital and get the treatment" (David, 15).

The participants did want to know more, however, about the procedures and the side effects of the medications: "[I'd like to know more about] the side effects [of] the medications. to know more about the treatment itself rather than the disease" (Edna, 16). They said that they would rather receive the full information than discover it by themselves: "They didn't **explain** to me when they inserted the port. I saw a video, but they didn't really **explain**" (Ruth, 13). "I didn't know about Ifosphamide [Chemotherapy drug]. I had side effects, but I didn't know I would get so thin. I'd like to know that in advance... to know all the side effects" (Eli, 18).

Only two participants spoke openly about the lack of knowledge concerning sexuality and intimate relationships given their illness, and their need for information and support: "I would like to know what I am allowed, what I am not allowed to do (with my girlfriend) – I asked my mother and she didn't know. I would like to know more in terms of my relationship. What's allowed? It feels like I have no one to talk to about it. Although I would like to talk about it" (Eli, 18).

Some Children and Adolescents Want to Get Any Kind of Information Even If It Is Intimidating, and Some Avoid It

The participants expressed no desire to know their prognosis or their chances of recovery, with one exception: "How did the illness develop? How many cases are there worldwide (because it is rare) and what is the chance of recurrence?" (Ron, 16).

The participants hardly talked about death and dying: "There are kids that may be scared by it (the information) and kids for whom it is a relief. For me, it's a kind of relief. At first, I thought you could die of it, but my condition was not bad. You can take care of yourself and stay alive. [My] parents explained it and it was reassuring" (Hope, 12). Only one participant talked about a life-threatening illness: "I know I have cancer in my blood. It's a life-threatening disease and it's a serious illness" (Avi, 16).

The majority of the participants (13/19) stated they do not need or do not want more information about their illness than they already know: "I'm not interested in knowing more"; "I wouldn't want to know more"; "It's not urgent for me. I get along as it is"; "I don't have to [know]. For me, it's enough."

The participants also talked about the negative effect of the information burden, which might be scary for children: "Too much information can cause fear" (Dan, 12). "They give the kids the least intimidating information. The scary information is not voiced. Some children want to know, and some don't want to know" (Johnny, 12).

Some participants spoke about the frightening sides of knowing and still said that they prefer to know because for them it was reassuring: "[On the other hand] it might be better not to know. Because sometimes when you know you're scared. And it can happen to you, just because of the thought about it. common side effects, but. . .. in spite of it, I prefer to know. I'd like to know more about what's going on" (Jonathan, 16). No differences in age-related information needs were found in the current study. A summary of the information known, the additional information the participants would like to receive, the information some of them would rather not know, and the requests of the participants can be seen in **Figure 1**.

The Opinions of Children and Adolescents About Truth-Telling

The participants spoke candidly about the merit of truth-telling to children. Esti (8.5), the youngest participant, wondered: "Why does the hair fall out? Why does the illness appear? And about the treatments and the pills. You need to say how long it will last; to tell children the truth from the beginning." Samuel (18), the oldest participant, added: "I feel like I am told everything, and it is important for me to know every step of the treatment, including future treatment options."

A few participants spoke about their feeling that information is being hidden from them: "At first, they didn't tell me anything. I was told that this is a kind of flu. Then I was hospitalized. I saw different children. And I didn't understand what this had to do with the flu. I inquired about it in more depth – What flu? What type of the flu? I asked the other kids what they were sick with. They told me it was blood cancer. If I could turn time back I'd like you to tell me the truth." (Eli, 18) "I think I know the large part of it. If there are other things? I believe they don't hide [things] from me" (Samuel, 18). Ron (16) said that he was satisfied with the information shared with him and stressed the importance of sharing: "Otherwise he/she might suspect that something is wrong." However, he said that the degree of sharing depends on the age of the child. Participants shared the reasons why they thought it was important to tell the truth. "I think a child should always be told the truth. A child who is not told can be shocked and angry that they are not included" (Jonathan, 16).

Ruth (13) added: "I don't like it when information is concealed from me. I want to be in control." She explained that she would rather not know about medications and leave it to her parents, but if anesthesia is involved, she needs to know what is being done, because when under anesthesia she is not in control. Avi (16) preferred "to know the truth, even if it is difficult to say; it is better [to hear it] directly than to seek the information from other sources. I searched the internet, Google. I don't think it's good to search on Google – most things are frustrating." He explained that he is an introverted person and the internet gave him the feeling that he had someone to talk to, but "even if a child is an introvert and does not ask for information, they should tell him." Most of the participants mentioned the importance of being included. One participant even mentioned the importance of the informed consent form for chemotherapy and the need to show it to adolescents even when under 18: "I only recently found out that there is such a form [informed consent]. . . I would have felt more comfortable if they had shown it to me much earlier. I think every adolescent should insist on reading it (Eli, 18)."

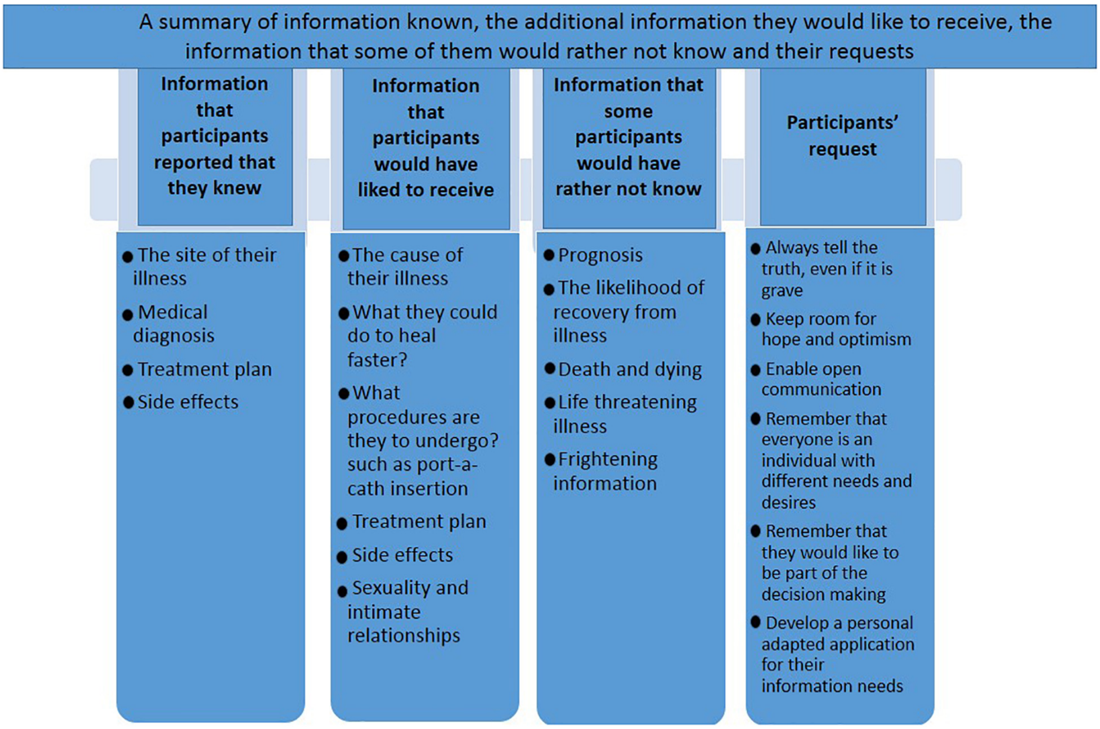
The Sources Children and Adolescents of Information and Support

As seen in **Table 3**, the participants identified several significant sources of information and support. The participants admitted that when they were diagnosed they had searched for information online. However, they no longer do so: "The internet was in the beginning. Today I ask doctors and nurses too" (Amir, 17); "I seek information from the doctors and nurses. I don't look for information on the internet because it's unreliable. It's better to ask people rather than a computer" (Avi, 16). Four adolescents claimed that it is important to have accurate information and that a customized app can fulfill this need: "I think we should build a structured and organized application that would provide us with all the details that are relevant for us" (Amir, 17). Avi (16) also thought of that idea: "We need to learn a new and complex language and the information needs to be tailored and relevant for everyone. Developing a special app can help us a lot. It is important that we have no question marks remaining, but we do not always know what to ask or feel comfortable asking."

Regarding the question from whom they received the information, 14 participants said that their primary source of information is physicians. Three participants said that their primary source of information is nurses and only two participants stated that their primary source of information is their parents.

Besides information as a helpful and supportive resource, the participants were asked what else helped them cope. Most of the participants noted the support of family and friends: "[What helps me are] conversations with dad, which gives me a view of more difficult situations" (David, 15). Two participants mentioned the good atmosphere in the [hospital] department as a source of support: "Always happy. The volunteers. raise morale" (Ron, 16); "[It is] always fun here [in the department]. So that's good for me." Esti (8.5) also mentioned that volunteers, friends, and family are those who most help cope with the disease.

When asked what helps them the most, the participants talked about how they maintain optimism, for example, Esti, (8.5) talked about the encouragement she receives from her surroundings: "The encouragement I get – that I'm good and I'm a champion." She also spoke about the thought that cancer would not last forever: ". . .the thought that everything will [eventually come to an] end. It strengthens me." Looking at cancer as a temporary situation was also how Amir (17) perceived it: "A child should know, remember, that it is only a [limited] period of time and that it will pass," and in the same spirit, Ron (16) described how practical knowledge has helped him remain optimistic: "Knowing at each stage what to do and why. Cancer is not the end of the world. You should stay optimistic." Samuel (18) revealed that he is encouraged by the thought of other children who are dealing with similar things and by those who have recovered from the disease: "To see other kids who are struggling



A summary of information known, the additional information they would like to receive, the information that some of them would rather not know and their requests

Information that participants reported that they knew	Information that participants would have liked to receive	Information that some participants would have rather not know	Participants' request
● The site of their illness ● Medical diagnosis ● Treatment plan ● Side effects	● The cause of their illness ● What they could do to heal faster? ● What procedures are they to undergo? such as port-a-cath insertion ● Treatment plan ● Side effects ● Sexuality and intimate relationships	● Prognosis ● The likelihood of recovery from illness ● Death and dying ● Life threatening illness ● Frightening information	● Always tell the truth, even if it is grave ● Keep room for hope and optimism ● Enable open communication ● Remember that everyone is an individual with different needs and desires ● Remember that they would like to be part of the decision making ● Develop a personal adapted application for their information needs

FIGURE 1 | A summary of information known, the additional information they would like to receive, the information that some of them would rather not known and their request.

TABLE 3 | The sources of information and support for children and adolescents.

Source of information	
Provider	**Interviewees' responses**
Internet	Was used only at the time of diagnosis Deemed as an unreliable source of information
Application	Non-existent. Was proposed as future development that could serve, as a suitable source of relevant information for them that could help them cope
Physicians	14 out of 19 participants perceived physicians as a primary source of information
Nurses	3 out of 19 participants perceived the nurses as a primary source of information
Parents	2 out of 19 participants perceived the parents as a primary source of information
Source of support	
Provider	**Interviewees' responses**
Family and friends	Most of the participants talked about the support that comes from family and friends and how much it strengthens them
Volunteers	The good morale in the hospital department and the volunteers who come and spend time with them help them cope

heroically." Edna (16) spoke about keeping her head up even in uncertain situations. She spoke about the need to be patient: "There is not always an immediate answer."

DISCUSSION

The current study adds knowledge received from adolescents with cancer regarding providing information about their illness. The analysis of the transcribed conversations with the research participants uncovered several important themes that recurred among most of them. In this chapter, we shall discuss these themes and compare them to previous findings in the literature.

The Need for Information of Children and Adolescents

Concerning knowledge about the disease, the participants knew the name of their disease and its site in their body and in this context, most did not feel that they need or would like to receive additional information. They were very interested in practical knowledge on the treatments *per se*, were familiar with the treatment protocol, and said that they would like to receive more information. The participants wanted to be more involved in decisions concerning the treatments and wanted the staff to see them as an inseparable part of the decision-making process. One participant even noted that, in his opinion, adolescents (under 18) should be asked to sign an informed consent form. During the interviews, the interpersonal diversity and complexity regarding the extent of the need for information about the treatments were conspicuous. The participants noted that receiving information helps them deal better with the disease and treatment protocol, however, excessive information is difficult for them and arouses fears and concerns. To meet the needs of the affected adolescents, the provision of information should be adapted to the individuals, their developmental stage, and their illness stage. Some said that information would lead to good and positive feelings such as confidence and trust in the medical staff and themselves, while for others, the same information would result in fear. Some claimed, furthermore, that even if certain information arouses concerns at an initial stage of the illness, at the end of the process, they see it as an advantage. The study shows that the participants saw the role of the medical staff as one that facilitates openness on various topics while maintaining sensitivity to the needs and wishes of the ill adolescent. One of the topics that occupy adolescents but was not freely expressed is related to intimacy, their sexual image, and sexual behavior. The two participants who initiated the conversation surrounding sexuality requested that the medical staff enable an open discourse on all possible topics, those that are easier to discuss but also those that are harder to talk about, such as sexuality. The difficulty to initiate conversations about sexuality is also supported by the study conducted by Veneroni et al. (2020).

What Information Is of Less Interest?

An interesting issue that the adolescents did not want to talk about was related to the prognosis. The participants said explicitly that such information might weigh heavily on them and prevent them from feeling hope. Perhaps not discussing their prognosis allows them to cope by disregarding and denying it so that they can hold on to hope. This clear message arose repeatedly in the interview with most of the participants. A study conducted by Mack et al. (2018) among adolescents aged 15–29 showed different results regarding the importance of conversations about prognosis issues from the perspective of the patients. Their study found that the patients who reported having received more extensive prognostic information experienced greater trust in the oncologist, greater peace of mind, and less distress. Despite the positive perception associated with the discourse on prognosis, their study found differences between the chances of recovery reported by the physician and the patient, where patients were inclined to perceive the chance of recovery in a more positive light than the physicians. It may be concluded that patients find it difficult to talk about a gloomy prediction regarding their future. This assumption can explain the current finding showing that adolescents did not want to talk about their prognosis. Adolescents may be so anxious about the negative outcomes of their illness may be reluctant to deal with threatening issues such as the risk of dying, that they prefer not to talk about them at all. Creative ways of enabling a protected discourse on these sensitive topics should be considered. The discussion of sensitive topics should begin with listening. The medical team should ask adolescents about their experience and listen to their answers before bringing up any feeling or explanation. In addition, they should offer honest answers but always leave room for hope and consider their needs, preferences, and condition.

Telling the Truth

During the interviews, the significance of honesty between the parents, medical staff, and the ill adolescent arose. Adolescents with cancer want information and want to know the truth. They sense when information is being concealed from them or when they are not included, and they become frustrated. The interviewees claimed that it is important to talk about anything the adolescent desires, and it is particularly important, to tell the truth, and not to conceal it, and certainly, not to lie. The participants suggested talking honestly about the situation even if it is grave. They claimed that the truth contributes to a sense of control even if it is hard to hear while concealing and not telling the truth lead to feelings of fear, loneliness, and distrust. It is evident from the findings that although everyone stated that the truth should be told, the age of the child has an important effect on how the truth is told, as it determines the determination of the child to deal with tough information. A 9-year old talked about the significance of telling the truth but claimed that words should be chosen carefully, while an 18-year-old emphasized that it is important for him to know everything – every stage of the treatment, and that he wants to be included in decisions that involve choosing between existing options. The need for age-adapted information is also supported by the literature review published by Raz et al. (2016) and Bibby et al. (2017). For example, it is important to choose the right words according to the age of the patient. Young children may better understand the meaning of the word “cancer” through images like “sick cells” or “soldiers in the body” as described by Esti (8.5) in the current study, while older children and adolescents may prefer to hear the words as they are such as “cancer,” “malignant,” etc.

Information Sources

Regarding information sources, the study showed that all the participants claimed that they would like to receive information from people – parents, doctors, and nurses. This information is important for them to feel involved and included. Similar to Coyne et al. (2016), in the current study, the participants stated that they trust their parents to provide them with the selected information that they should know. The parents constitute their anchor and support system and they appreciate them as the mediators of information who adapt it for their children so that they will be able to deal with it. A meta-analysis conducted by Yamaji et al. (2020) revealed similar findings, which highlighted that children perceived their parents as reliable suppliers of information and as an important support resource.

Beyond the parents as information suppliers and to confirm the information they possess, adolescents also ask the staff. This finding is consistent with other studies showing that health care providers are perceived as the primary source of information for AYA (Kent et al., 2016). Despite the wish of the adolescents to receive information from nurses, the interviews show that although the nurses are in close and continuous contact with the adolescents and their families, they are less perceived by adolescents as the main information resource.

Only a few of the interviewees used the internet as a source of information, and they did so, particularly, in the initial stage. The interviewees said that in the course of their illness, the further they got from their diagnosis, they avoided searching for information on the internet both because they received the information they need from significant others (parents and staff) and because the information on the internet is general and is not tailored to their specific state or condition which might lead to feelings of fear, insecurity, and uncertainty. Even participants who would search for information about their disease on the internet immediately after diagnosis quickly realized that this information was too general, did not suit them personally, and was sometimes threatening and harsh. Research conducted among cancer survivors aged 18–39 years revealed similar findings regarding the vast and extensive information available on the internet. The participants who searched for healthy lifestyle information on the internet said that there was too much information on the internet and that the information they found was not tailored to their unique challenges and needs as AYA cancer survivors. One AYA cancer survivor stated, "Everybody is different, so there would need to be some specialized programs based on your diagnosis and what's recommended" (Mooney et al., 2017).

The current study shows the need for advanced technology in the service of information that is personally adapted to the patients. A similar finding arose in a Canadian study held among 33 adolescents, aged 12–18 years, with chronic illnesses. The participants claimed, similar to the findings of the current study, that they are interested in developing a personally adapted application that would help them cope and give them access to trustworthy, professional, and authorized information (Kohut et al., 2017).

Clinical Implications

This study includes a sample of 19 eligible Israeli adolescents who completed a psychological interview consisting of 15 open-ended questions used in previous studies. The main results showed that whereas the participants knew enough about the course of their illness, they were not informed about its etiology. They were very interested in practical knowledge about the treatments *per se*, were familiar with the treatment protocol, and would like to receive more information about the treatments and sexuality and intimacy. The participants reported a lack of knowledge regarding these issues and felt a sense of discomfort talking about them. The adolescents would like the medical professionals to leave them with no question marks and to provide them with the information they need, especially because they do not want to use the internet as a source of information due to the profusion of stressful and inadequate information it contains. The findings also revealed that adolescents want to receive information and to be involved in decision making and yet want the information they receive to be positive and to leave them with optimism and hope for the future.

The study indicates that similar to that known from the literature, adolescents are indeed a unique population that needs special attention and special understanding due to the many physical, mental, and social changes typical in adolescence, together with the traumatic crisis surrounding a life-endangering, scary, and lengthy illness. The knowledge, understanding, and open and adapted channels of communication concerning the disease and its treatment between the adolescent, the family, and the medical staff, are crucial for the ability of the adolescent and their family to handle the hardships caused by the treatment, the illness, and the recovery process. The research findings help the staff understand the communication and information needs of adolescents who are at a complex stage of their development, between childhood and maturity. They are no longer children but have not yet matured as adults. At this stage of their development, they seem to require a balanced combination of the parental protection they need as children and the desire to know, share, and control what they need as adults. The adolescents seek to be involved but also seek a type of protection during conversations related to their medical condition. They need to be part of a positive discourse that will leave room for hope. It is important for them to hear, as shown by Jalmsell et al. (2016), that the illness is a passing phase in their life, and that the future awaits them.

The following recommendations were derived from the contents raised by the interviewees in the study. These recommendations might advance treatment, as well as provide access to information for adolescents with cancer:

1. The medical staff must include the patient and actively provide access to information, while demonstrating honesty and sensitivity.
2. Initiated conversations can take place even when not actively requested by the adolescents, to provide them with information that is as inclusive and accurate as possible.
3. The issue of age and interpersonal differences that lead to different desires for information by adolescents should be taken into account before providing information. It is desirable to examine and explore whether there are topics that they are afraid or ashamed to talk about.
4. Before initiating a conversation, it would be desirable to ask: "What do you already know and what would you like to know more about?"
5. Presenting an informed consent form for chemotherapy to patients should be considered even in the case of adolescents who have not yet reached the legal age of informed consent. Full inclusion might contribute to a sense of belonging and involvement.
6. Finding the resources to develop a personally adapted application that adolescents can use to detect where they are on the health and illness sequence and to receive accurate information when desired, should be considered. This can involve developing a type of "Onco-Waze" (road map). This application might reduce concerns and

uncertainty and increase knowledge and a sense of security and partnership.

7. Parents should be instructed, from the stage of diagnosis, to share adapted information and to avoid saying anything that does not arouse trust.
8. It must be clarified to parents that adolescents are capable of dealing with tough information positively and that they prefer this over dealing with a lack of information or incorrect information.
9. A supportive environment contributes to positive feelings among adolescents. Try to produce and encourage social support through friends, family, and volunteers.
10. Staff should be trained to care for the population of adolescents with cancer by providing knowledge about this complex developmental stage, in times of health and sickness. By training the staff, it is possible to provide a more adjusted response for this population and thus, help them cope during the treatment period and subsequently when resuming their routine.
11. Similar studies should be conducted to facilitate evidence-based research on how information is provided and the type of information that is suitable for adolescents with cancer.

Research Limitations

The research findings should be interpreted taking into account several limitations. One is related to the small sample size, which may be a limitation on the conclusion. However, it should be noted that childhood cancer is a rare disease and of all children with cancer, adolescents constitute an even smaller percentage. In addition, the age and gender of the research participants were heterogeneous, and in each age group, there was a limited number of interviewees. This may have inhibited finding significant differences for each age and gender group with regards to the information needs or different perceptions of adolescents regarding the issues that arose in the study. Another limitation is related to the religiosity of the interviewees. Most of the research participants defined themselves as religious, and this can affect the information-related issues that occupy them. The religiosity of the participants might explain why only two interviewees (who were secular) raised the need for information on sexuality. The nationality of the interviewees may constitute a further limitation of the study. All the interviewees belonged to a single nationality. Further research that will also include sick Muslim and Christian participants on different levels of religiosity may be able to examine cultural differences in the information needs of adolescents with cancer in Israel. Another limitation is related to the fact that the participants were treated at a single hospital in Central Israel. Examining the information needs of adolescents treated at other hospitals would provide a more comprehensive picture.

CONCLUSION

In conclusion, adolescents with cancer need trustworthy information and prefer to receive it from a human source rather than from the internet, such as Google, etc. Not being told the truth can arouse negative feelings of fear and loneliness in adolescents. Thus, medical professionals should operate in sensitive ways to provide adolescents with access to information on various subjects, including sexuality. Open communication and trust relations between the medical staff, parents, and adolescents may be a key to enhancing the resilience and well-being of adolescents (Mack et al., 2018; Yamaji et al., 2020). Besides human sources of information, there is room to consider developing technologies that will provide adolescents with personalized information appropriate for their needs, which will be available to them in any situation and at any time.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding authors.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Helsinki Committee at the Rabin Medical Center. Written informed consent to participate in the study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

All authors listed have made a substantial, direct and intellectual contribution to the work, and approved it for publication.

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Relation Between Social Support Received and Provided by Parents of Children, Adolescents and Young Adults With Cancer and Stress Levels and Life and Family Satisfaction

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Introduction: The present study aims at analysing how social support received and provided by parents of children, adolescents and young adults (AYA) diagnosed with cancer, as well as their sociodemographic and clinical variables, affect those parents' stress levels and life and family satisfaction.

Materials and Methods: A total of 112 parents of children and AYAs who had been diagnosed with cancer and who received treatment in Malaga participated in the study. In the study, participated all parents who voluntarily agreed to fulfil the questionnaire. The main inclusion criterion was that their child had cancer. Instruments used were Questionnaire on the Frequency of and Satisfaction with Social Support (QFSSS), Paediatric Inventory for Parents (PIP), Life Satisfaction Scale and Family Satisfaction Scale.

Results: In the mean difference analyses, male parents showed 3.38 (SD = 0.56) in social support received and female parents showed $M = 3.08$ (SD = 0.72). Conversely, in social support provided, female parents showed 3.22 and male parents showed $M = 3.55$ ($p = 0.020$). Significant differences were also found in family satisfaction, where female parents ($M = 3.64$) feel more satisfied than male parents ($M = 3.06$; $p = 0.027$). Parents of children aged between 0 and 14 years ($M = 3.06$) feel more stress than those parents of children aged 15–21 ($M = 2.61$; $p = 0.021$). The correlation analysis shows that there is a negative and significant relation between stress levels experienced by parents when facing different situations related to the child's disease and both types of support, received $r = -0.411$, $p < 0.001$ and provided $r = -0.282$, $p < 0.01$. There is also a positive and significant relation between life satisfaction and social support received $r = 0.292$, $p < 0.01$, and social support provided $r = 0.409$, $p < 0.001$. There is a positive and significant relation between family satisfaction and social support received $r = 0.330$, $p < 0.01$, in the same way as with social support provided $r = 0.222$, $p < 0.05$. The regression analysis related to stress levels of parents indicates that social support received predicts levels of stress significantly $p < 0.001$, with the variable of number of children being the one that showed to be significant $p < 0.05$. Social support provided showed the most significant results $p = 0.001$, meaning that social support provided increased life satisfaction. Social support received explains family satisfaction ($p = 0.50$), as it increases the family satisfaction of parents of children with cancer.

Discussion: Analysing social support received and provided, as well as sociodemographic and clinical variables, allowed us to broaden the knowledge on the effect social support has on stress levels, life satisfaction and family satisfaction in parents of children and AYAs diagnosed with cancer. This may have relevant practical implications for the design of interventions that would improve parents' lives.

Keywords: social support received, social support provided, parents of children with cancer, stress levels, family satisfaction, life satisfaction

INTRODUCTION

Cancer is a disease that affects the lives of patients and their relatives both socially and psychologically (Molinaro and Fletcher, 2018; Carlsen et al., 2019; Wilford et al., 2019) and it is a great challenge for those families whose members diagnosed with cancer are minor CHILDREN OR ADOLESCENTS (Van Schoors et al., 2017; Sánchez-Egea et al., 2019). We refer to childhood cancer that includes ages 0 to 14. In this age range, minors are treated by a specialised service of paediatric oncology. In recent years, reference has begun to be made to childhood-teenage cancer, including the ages of 15 to 21. Traditionally, patients within this age group would receive adult oncology services; however, increasingly more units, services and hospitals are considering adolescents and young adults (AYA) between 15 and 21 to be closer to the paediatric oncology approach, thus understanding them with their own characteristics and particularities. This approach also aims at applying a more appropriate transition from teenage to young adulthood and hence provide better services to these groups (Ministerio de Sanidad, Servicios Sociales e Igualdad de España, 2015). Families facing childhood cancer must deal with a wide range of situations (Enskär et al., 2020), which include high frequency of treatments and hospitalisation of the minor, side effects associated with such treatments, uncertainty about the course of the disease and fear of a possible relapse (Sloper, 2000; Ångström-Brännström et al., 2018). These situations contribute to higher levels of stress in parents (Hoven et al., 2017; Tillery et al., 2020). In fact, many parents have shown symptomatology linked to stress even after their children overcome the disease (Norberg and Boman, 2013; Bilodeau et al., 2018). The situations linked to most of the stress parents suffer are related to hospitalisations (Liu et al., 2020), receiving information related to the child (Smith et al., 2019) and waiting times in diagnoses and tests (Patterson et al., 2003).

There are some protective factors against stress. One of the most relevant is receiving social support, which makes it possible for parents not to experience the same psychological effects over time (Schirren and Boman, 2010). The study of social support is becoming a field of special interest in psycho-oncology (Herrero and Gracia, 2007; Gunter and Duke, 2019), as it provides information on how patients and their relatives cope with the disease. More precisely, social support has proved to reduce stress levels in patients (Sloper, 2000). Support is understood as an exchange of aid provided by one person to another, as well as the social resources that individuals perceive as available at a given time (Gottlieb and Bergen, 2010). Support relates to feeling valued and cared for as part of a social network of mutual support (Taylor et al., 2004).

Very few studies analyse social support taking into consideration its double dimension, that is, satisfaction with social support received and satisfaction with social support provided to others. The same person may provide support and receive it at the same time. It is therefore important to know the impact of this double function of support in variables, such as stress and life and family satisfaction of parents of children with cancer (Sloper, 2000; Carlsen et al., 2019). Social support should be analysed in a multidimensional way (Melguizo-Garín et al., 2019); however, most studies only analyse the social support that parents receive from their networks. Studies like the one by Tremolada et al. (2012) show that the perception of social support that mothers of children with leukaemia receive relates to less psychological symptoms and higher satisfaction with life. Other authors suggest that perceiving support could be related to feeling less distress caused by the child's diagnosis (Hoekstra-Weebers et al., 2001). Jou and Fukada (2002) note that for support to be beneficial, there must be a balance between what is given and what is received. When individuals provide more support than they receive, they may feel an excessive burden. Conversely, when individuals receive more support than they provide, self-esteem decreases and there is a feeling of being a debt. Jaeckel et al. (2012) also found that the lack of reciprocity in social support could have negative effects on wellbeing. Despite this dimension of social support provided having been scarcely studied, the balance between support received and support provided is key for the life satisfaction and health of individuals as it is shown by the studies mentioned above.

Cancer has a significant impact on life satisfaction and parents' family satisfaction. Parents' satisfaction is one of the variables that is most affected in the process of childhood cancer (Faith et al., 2019). Different situations related to the child's treatment and the alteration of family routines are sources of stress and can also lead to low life satisfaction (Espada and Grau, 2012). Social support has a positive effect on parents' life satisfaction (Gibbins et al., 2012; Marsland et al., 2013). Another area that has been proved to be affected in parents of children with cancer is family satisfaction. When childhood cancer appears, parents focus on caring for the ill child, which can have a negative impact on family life, family satisfaction and the quality of family relations (Salvador et al., 2019). This notion is deeply determined by individual, relational and social factors (Acevedo et al., 2007). Family satisfaction is understood as the interactional wellbeing of the family members (Sobrino, 2008). Low family satisfaction resulting from family difficulties due to the child's disease is related to lower quality of life (Salvador et al., 2019). Cohesion between family members is tightly linked to family satisfaction and positive adaptation to the situation is negatively linked to stress suffered by parents (Yoon et al., 2018). Authors, such as Wasserman and Danforth (1988), consider that social support constitutes a family phenomenon and a shared place of interaction between individuals. It seems that family can be considered as a space where its members relate spontaneously with one another and in which non-problematic events as well as those events that jeopardise the family balance must be dealt with. There is little research on how social support relates to the family satisfaction of those parents whose child has cancer, even more if we consider the double functionality of social support (received and provided).

The present study aims at analysing how receiving and providing support by parents of children with cancer affects stress levels and life and family satisfaction. The variable of family satisfaction in the context of childhood cancer has been scarcely studied. One of the new contributions of this study is that social support is analysed considering two dimensions, social support received by parents of children with cancer and social support provided by them (to their network and close family). There is little research on the effect of social support received and provided in the context of families with children with cancer and its relation to life and family satisfaction, as well as stress levels.

Considering the objectives of the study, the following hypotheses are suggested as:

1. There is a negative relation between satisfaction from social support received and provided and stress levels.
2. There is a positive relation between satisfaction from social support received and provided and life and family satisfaction.
3. Higher levels of satisfaction from social support received and provided predict lower levels of stress and higher life and family satisfaction.

MATERIALS AND METHODS

Participants

A total of 112 parents of children and AYAs with cancer who received treatment at the Children's Hospital of Málaga (Spain) participated in the study. Participants were selected based on their voluntary wish to participate. Participants were at different stages of their children's cancer disease and treatment. The sample was gathered from parents of children and adolescents who received treatment at the Mother and Child Hospital of Malaga and from parents of patients aged between 15 and 21 from the Regional Hospital of Malaga (Spain). Once parents were explained the aim of the study and what the procedure would be like, those who decided to participate in the study formed the final sample. Inclusion criteria for the study sample were the following: parents or legal guardians of patients aged from 0 to 21 with cancer disease. Conversely, exclusion criteria were the following: other relatives of patients who were not the parents or legal guardians and parents whose child had deceased. The sociodemographic questionnaire did not include questions about the stage of treatment (on-going or follow-up). However, all participants attended the Hospital for their children to receive treatment related to cancer (follow-up consultations, ambulatory treatment, hospitalisation, etc.).

Procedure

Parents who participated in the study went regularly to the Children's Hospital of Malaga. They were contacted directly at the hospital (oncology rooms, semi—private accommodation and hospitalisation area) or in some rooms for children with cancer that the local NGO has within the hospital used to provide support to families of children with cancer. Participants received a written and verbal informed consent about the procedure that would be carried out. The study was approved by the Ethical Committee on Scientific Research from the Regional Government of Andalusia (Spain), CEI 2017. Once participants had signed the informed consent, they were given the option to choose between two ways of completing the instrument: on paper during some of their visits to the hospital or online through a digitalized version of the instrument, which was anonymously and automatically added to a database upon completion.

Measures

Sociodemographic Questionnaire

The sociodemographic questionnaire included questions related to gender, age and marital status of participants (level of qualifications, employment situation, number of children and number of people under their care). This questionnaire also includes questions about the child: gender, type of cancer and length of time since diagnosis.

Questionnaire on the Frequency of and Satisfaction With Social Support

Questionnaire on the Frequency of and Satisfaction with Social Support (QFSSS) by García-Martín et al. (2016) was used. This questionnaire was used to measure social support received and provided by parents. More precisely, we measured the type of support (emotional, instrumental and informative) provided by the different sources of support from parents' social networks (partners, relatives, friends and members of the community and the association), as well as the type of support provided by parents to the different sources. The questionnaire comprises 12 items on support received and 12 items on support provided. Answer options for each dimension are five, where '1' means never and '5' means always in terms of frequency and '1' means unsatisfied and '5' very satisfied in terms of satisfaction. This study uses the average score of social support received considering the three types of support and the four types of sources, as well as the average score of social support provided considering the same three types and four sources of support. Cronbach's Alpha of the full scale is $\alpha = 0.96$. The following are some examples of the items included in the satisfaction with social support received section: '*you receive care and affection and you are listened to when you need to talk and express emotions* (emotional support from partner) and '*He/she is willing to do things for you, such as helping you with your daily chores or in the care of your child*' (functional support), and these are some examples of items included in the satisfaction with social support provided section: '*you give them useful advice and information to solve their doubts, problems or daily chores*' (informational support provided to friends) and '*You are willing to do specific things for them, such as helping them with their daily chores or care*' (informational support provided to community).

Paediatric Inventory for Parents

Paediatric Inventory for Parents (PIP), by Streisand et al. (2001), was used, in its adaptation and validation from Del Rincón et al. (2007), Spanish version. This questionnaire measures levels of stress caused by situations parents of children with cancer face daily. It comprises a frequency scale and an effort scale. The following are two examples of items included in the frequency scale and the effort scale, respectively: '*How often do you experience sleeping problems?*' and '*How difficult is it for you to attend your child's medical tests and treatments*'. Each scale comprises 42 questions related to situations faced by parents during their child's disease. Participants must answer how frequently each situation occurs and must choose between five options, where '1' means never and '5' means very often. Subsequently, they must also answer according to the effort such situations take, where '1' means none and '5' means very much. Cronbach's Alpha of the full scale is $\alpha = 0.95$.

Life Satisfaction Scale

Life Satisfaction Scale from Pavot and Diener (1993) was used. This scale offers a general index of life satisfaction, where life satisfaction is understood as a general construct of subjective wellbeing. It is a unidimensional scale comprising five items, which are answered through a Likert-type scale of 7 points (1 = completely disagree and 7 = completely agree). The following is and an example of an item in this scale: '*In most things, my life is very close to how I want it to be*'. Cronbach's Alpha of the full scale is $\alpha = 0.87$.

Family Satisfaction Scale

Family Satisfaction Scale from Olson and Wilson (1982) was used, in its translated and adapted version. The scale was reduced to 10 items (Olson and Wilson, 1982), which measure the level of satisfaction in relation to family cohesion and adaptability. Answers to the Family Satisfaction Scale range between '1' (very unsatisfied) and '5' (very satisfied). This study uses the total average score obtained by calculating the average of all scores and then unifying both dimensions, family cohesion and adaptability. The following are some examples of items included in this questionnaire: '*My family's ability to share positive experiences*' and '*the quality of the communication between the family members*'. Cronbach's Alpha of the full scale is $\alpha = 0.95$.

Data Analyses

Statistical analyses were carried out through SPSS (v.25). No missing values were found. Data met the criteria to carry out the suggested analyses and there was no collinearity between the study's variables. First, sociodemographic and clinical characteristics of the study's sample were analysed, and then, a descriptive analysis of the study's variables was carried out. Mean differences were also analysed to verify if there were significant differences between results of the study's variables in the sample based on parents' gender, children's and AYAs' age and time from diagnosis. In order to know the relation between the different variables (clinical and sociodemographic variables of the sample, social support received and provided, stress and life and family satisfaction), the Pearson's correlation coefficient was calculated. Finally, a multiple linear regression analysis was carried out to find out more about the existing relations between the variables of the study. Three regression models were carried out as: one for stress, another for life satisfaction and a third one for family satisfaction. These variables acted as dependent variables. Independent variables in the three models suggested were as: social support received and social support provided, and the sociodemographic and clinical variables of the sample. The variables considered for the regression equation were those with statistical relevance ($p < 0.05$).

RESULTS

Demographic and Clinical Characteristics of the Study Cohort

The sample comprised 33.9% men and 66.1% women, with an average age of 41 years (SD = 6.93). Most participants were married or lived with their partners (88.5%). The remaining were single, divorced or widowed (11.5%). Regarding the gender of the children, 58% were boys and 42% girls, with an average age of 8 years (SD = 5.02). Regarding the type of cancer, 54.5%

suffered from leukaemia, 9% from Ewing sarcoma, 8% from lymphoma, 4.5% from medulloblastoma and the remaining suffered from other types of childhood cancer. The length of time from diagnose was the following: 18.9% of children had been diagnosed less than 1 year before, 23.4% 1 year, 19.8% 2 years ago, 9.9% 3 years ago, 12.6% 4 years ago and the remaining 15.3% 5 or more years ago. All data can be seen in **Table 1**.

TABLE 1 | Sociodemographic variables ($n = 112$).

Parents	
Age of parent/guardian	41(6.93)[a]
Number of children	1.98(0.67)[a]
Number of people under care*	2.18(1.15)[a]
Age of child with cancer	8(5.02)[a]
	%(*N*)
Age of child 0–14 years	87.5(98)
Age of child 15–21 years	12.5(14)
Gender of the parent/guardian	
Male	33.9(38)
Female	66.1(74)
Marital Status	
Single	3.6(4)
Married	80.4(90)
Divorced	3.6(4)
Separated	3.6(4)
Widowed	0.9(1)
Unmarried partner	1.8(2)
Living as a couple	6.3(7)
Education Level	
University degree	31.3(35)
Vocational Training	33(37)
A Levels	11.6(13)
Secondary Education	22.3(25)
Other	1.8(2)
Employment Situation	
Civil Servant	13.4(15)
Self-employed	11.6(13)
Employee	31.3(35)
Unemployed	33(37)
Domestic work	10.7(12)
Children	
Gender of the child with cancer	
Boy	58(65)
Girl	42(47)
Type of cancer	
Leukaemia	53.6(60)
Ewing Sarcoma	8.9(10)
Lymphoma	8(9)
Medulloblastoma	4.5(5)
Neuroblastoma	4.5(5)
Rhabdomyosarcoma	2.7(3)
Hepatoblastoma	2.7(3)
Astrocytoma	1.8(2)
Other	13.4(15)
Length of time since diagnose	
Less than 1 year	18.9(22)
1 year	23.4(26)
2 years	19.8(22)
3 years	9.9(11)
4 years	12.6(14)
5 or more years	15.3(17)

[a] *Average (Deviation).*
**Carers of the elderly, dependent persons.*

Descriptive Characteristics of the Outcomes

To know participants' stress levels, their social support received and provided and their life and family satisfaction, descriptive analyses were carried out, as it can be seen on **Table 2**. Results indicate that parents express feeling medium-high levels of stress when they must face situations related to their child's disease $M = 3.01$, SD = 0.67. They report receiving medium levels of support $M = 3.18$, SD = 0.68 and providing medium-high support $M = 3.33$, SD = 0.70. They also express having medium-low levels of life satisfaction $M = 3.52$, SD = 1.17 and medium-high levels of family satisfaction $M = 3.48$, SD = 0.90. The reliability of each instrument used in this study can be observed in **Table 2**. This was calculated through Cronbach's alpha.

Mean Differences

To find out whether there are significant differences between parents according to gender, the age of their ill children (and if these children belonged to the group 0–14 years or 15–21 years) and according to the time passed from the diagnosis, *T*-tests on independent samples were carried out. Through these tests, the aim was to know the differences in the variables of social support received, social support provided, stress, life satisfaction and family satisfaction. Results from analyses are shown below.

The *T*-test with participants' gender as grouping variable showed significant differences when it comes to satisfaction with social support received and social support provided. For social support received, male parents show a mean of 3.38 (SD = 0.56) and female parents $M = 3.08$ (SD = 0.72) with $p = 0.015$. Regarding social support provided, female parents show a mean of 3.22 (SD = 0.73) and male parents $M = 3.55$ (SD = 0.61) with $p = 0.020$. This means that fathers feel more satisfied with the support they provide and receive than mothers. Significant differences were also found in family satisfaction, where female parents showed $M = 3.64$ (SD = 0.84), thus meaning they feel higher family satisfaction than male parents $M = 3.06$ (SD = 0.96) with $p = 0.027$. Regarding stress and life satisfaction, no significant differences were found based on participants' gender (**Table 3**).

The *T*-test with children's age as grouping variable showed significant differences according to the stress variable. Parents of children aged between 0 and 14 $M = 3.06$ (SD = 0.66) experience more stress than those parents whose children are aged between 15 and 21 years $M = 2.61$ (SD = 0.68) with $p = 0.021$. Regarding the remaining variables (social support

TABLE 2 | Mean and standard deviation of social support received and provided, stress and life and family satisfaction.

	N	*M*	SD	*Reliability*
Support received	112	3.18	0.68	0.92
Support provided	112	3.33	0.70	0.95
Stress	112	3.01	0.67	0.95
Life satisfaction	112	3.52	1.17	0.87
Family satisfaction	112	3.48	0.90	0.95

TABLE 3 | Mean differences in social support, stress, life satisfaction and family satisfaction according to the gender of the parent.

	Social support received		Social support provided		Stress		Life satisfaction		Family satisfaction	
	***M*(SD)**	***p***	***M*(SD)**	***p***	***M*(SD)**	***p***	***M*(SD)**	***p***	***M*(SD)**	***p***
Gender										
Male	3.38(0.56)	0.015	3.55(0.61)	0.020	2.85(0.68)	0.076	3.80(0.99)	0.081	3.06(0.96)	0.027
Female	3.08(0.72)		3.22(0.73)		3.09(0.66)		3.39(1.24)		3.64(0.84)	

TABLE 4 | Mean differences in social support, stress, life satisfaction and family satisfaction according to the age range of the child.

	Social support received		Social support provided		Stress		Life satisfaction		Family satisfaction	
	***M*(SD)**	***p***	***M*(SD)**	***p***	***M*(SD)**	***p***	***M*(SD)**	***p***	***M*(SD)**	***p***
Age range of the child										
0–14	3.19(0.68)	0.677	3.34(0.71)	0.628	3.06(0.66)	0.021	3.52(1.20)	0.991	3.52(0.93)	0.390
15-over	3.11(0.70)		3.25(0.64)		2.61(0.68)		3.53(1.01)		3.18(0.54)	

TABLE 5 | Mean differences in social support, stress, life satisfaction and family satisfaction according to the diagnosis time.

	Social support received		Social support provided		Stress		Life satisfaction		Family satisfaction	
	***M*(SD)**	***p***	***M*(SD)**	***p***	***M*(SD)**	***p***	***M*(SD)**	***p***	***M*(SD)**	***p***
Diagnosis time										
0–2 years	3.27(0.63)	0.102	3.30(0.65)	0.494	3.00(0.64)	0.891	3.66(1.10)	0.123	3.49(0.84)	0.971
3-over years	3.05(0.75)		3.40(0.79)		3.02(0.74)		3.30(1.28)		3.48(1.03)	

received, social support provided, life satisfaction and family satisfaction), no significant differences were found based on children's ages (**Table 4**).

Lastly, a *T*-test was carried out with time since diagnosis as grouping variable. As it can be seen on **Table 5**, no significant differences were found when testing this variable with the study's variables (social support received, social support provided, stress, life satisfaction and family satisfaction).

Univariate Analysis

Existing relations between support received and provided by parents and stress and life and family satisfaction are described in this section. Pearson's correlation coefficients were calculated, as it can be seen in **Table 6**. Results show there is a negative and significant relation between stress levels experienced by parents when facing different situations related to the child's disease and both types of support, received $r = -0411$, $p < 0.001$ and provided $r = -0.282$, $p < 0.01$. There is also a positive and significant relation between life satisfaction and social support received $r = 0.292$, $p < 0.01$, and social support provided $r = 0.409$, $p < 0.001$. Likewise, a positive and significant relation was also found between family satisfaction and social support received ($r = 0.330$, $p < 0.01$), and social support provided ($r = 0.222$, $p < 0.05$.). No significant relations were found between sociodemographic and clinical variables and stress, life satisfaction and family satisfaction.

TABLE 6 | Correlations between social support received and provided and clinical and sociodemographic variables with stress, life and family satisfaction.

	Stress	Life Satisfaction	Family Satisfaction
Social support received	−0.411***	0.292**	0.330**
Social support provided	−0.282**	0.409***	0.222*
Age of parent/guardian	−0.112	0.002	−0.305*
Number of children	−0.188	0.055	0.124
Age of child with cancer	−0.059	−0.079	−0.231
Diagnosis time	−0.101	−0.008	−0.063

$*p < 0.05$; $**p < 0.01$; and $***p < 0.001$.

Multivariable Analysis

A multiple linear regression analysis was carried out on three models suggested. Models can be seen further down. Social support received and provided and the sociodemographic and clinical variables were considered independent variables in each model; dependent variables were stress, life satisfaction and family satisfaction.

For the first model analysed, parents' stress was considered dependent variable (**Figure 1**). The regression analysis related to stress levels of parents indicates that social support received predicts levels of stress significantly $p < 0.001$, meaning that social support received reduces parents' stress levels (**Table 7**). Social support provided did not show significant results. When

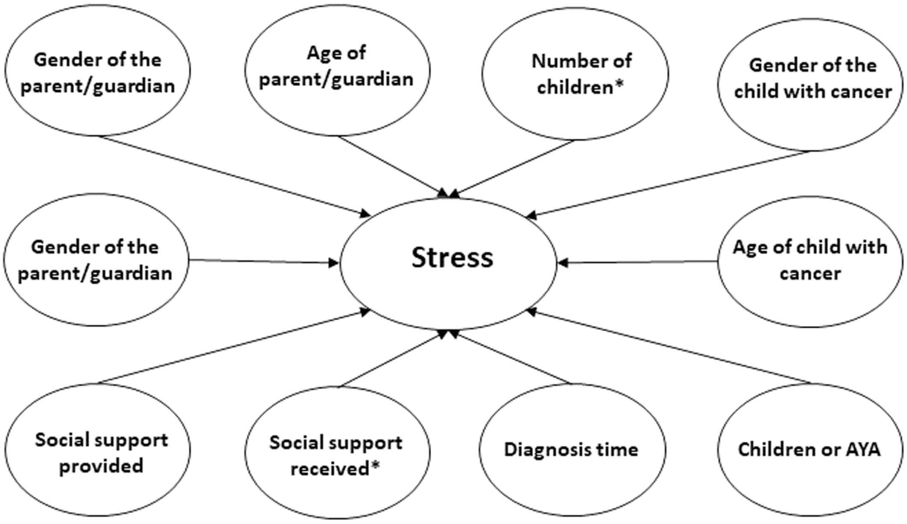

FIGURE 1 | Stress model.

analysed according to sociodemographic and clinical variables as independent in the model, only the number of children was found to be significant $p < 0.05$. The size of the effect of social support received and the number of children over stress is low ($R^2 = 0.28$), meaning that 28% of the variance is explained by the social support that parents receive and the number of children they have.

In the second model analysed, life satisfaction was considered dependent variable and social support received and provided and sociodemographic and clinical variables were considered independent (**Figure 2**).

In this case, social support provided showed the most significant results $p = 0.001$, meaning that social support provided increased life satisfaction. The size of the effect of the model is $R^2 = 0.22$, meaning that 22% of the variance is explained by social support provided. Social support received and the remaining sociodemographic and clinical variables did not predict life satisfaction. These results can be seen in **Table 7**.

In the third model, family satisfaction was considered dependent variable and social support received and provided and sociodemographic and clinical variables were considered independent (**Figure 3**). Social support received explains family satisfaction ($p = 0.50$), as it increases the family satisfaction of parents of children with cancer. The size of the effect of social support received on family satisfaction is $R^2 = 0.30$, el 30% de la varianza es explicada por el apoyo social recibido. Social support provided and the remaining sociodemographic and clinical variables were not statistically significant (**Table 7**).

DISCUSSION

Most relevant results from this research show that male parents feel more satisfied with social support received and provided; however, it is female parents who feel higher family satisfaction. Parents of children aged 0–14 feel more stress than those parents of children aged 15–21. A positive and significant relation was found between satisfaction with social support received and provided and life and family satisfaction. However, a negative and significant relation between social support received and provided and stress was found. Additionally, social support received and parents' number of children predict stress in parents, where higher satisfaction with social support received and higher number of children relate to lower levels of stress. On the other hand, social support provided by parents is the one that relates to life satisfaction, where higher satisfaction with social support provided shows higher life satisfaction. Finally, satisfaction with social support received relates to higher levels of family satisfaction.

Analyses conveyed emphasise the importance of knowing how the two dimensions of social support and sociodemographic and clinical variables affect stress levels as well as life and family satisfaction in parents of children with cancer. Most research on social support mainly focuses on analysing social support received. More specifically, studies have shown that support is a source that helps parents cope with the difficult situations caused by childhood cancer (Haunberger et al., 2019). However, social support provided by parents to other people facing the same situation is also a key variable to cope with such a difficult situation, as confirmed by the results from the present study.

In general, the hypotheses suggested were confirmed. The significant and negative relation between the two dimensions of social support and stress seems rather clear, as well as the significant and positive relation between social support received and provided and life and family satisfaction. When the effect of social support received and provided on the studied variables is analysed more in-depth results obtained are worth noting. Social support received is the variable that better predicts a decrease in stress levels, as well as higher family satisfaction.

TABLE 7 | Multiple linear regression analysis for stress, life and family satisfaction ($n = 112$).

Model	Non-standardised coefficients		Standardised coefficients	*t*	*p*
	B	***Standard error***	***Beta***		
Stress					
(constant)	4.854	0.642		7.563	0.000**
Gender of the parent/guardian	0.113	0.128	0.080	0.886	0.378
Age of parent/guardian	−0.004	0.011	−0.043	−0.375	0.709
Number of children	−0.200	0.096	−0.189	−2.089	0.039*
Gender of the child with cancer	0.216	0.123	0.160	1.754	0.083
Age of child with cancer	0.002	0.019	0.015	0.110	0.913
Children or AYAs	−0.433	0.244	−0.211	−1.775	0.079
Diagnosis time	−0.014	0.127	−0.010	−0.107	0.915
Social support received	−0.372	0.116	−0.375	−3.198	0.002**
Social support provided	−0.028	0.109	−0.029	−0.254	0.800
$R = 0.528$, $R^2 = 0.279$, R^2 adjusted $= 0.212$, $F = 4.176$, Sig = 0**					
Life satisfaction					
(constant)	2.244	1.169		1.920	0.058
Gender of the parent/guardian	−0.202	0.231	−0.081	−0.871	0.386
Age of parent/guardian	0.000	0.020	0.001	0.010	0.992
Number of children	0.086	0.162	0.049	0.534	0.595
Gender of the child with cancer	−0.271	0.221	−0.115	−1.228	0.223
Age of child with cancer	−0.006	0.035	−0.026	−0.177	0.860
Children or AYAs	0.159	0.447	0.044	0.357	0.722
Diagnosis time	−0.404	0.230	−0.166	−1.759	0.082
Social support received	−0.017	0.212	−0.009	−0.078	0.938
Social support provided	0.693	0.197	0.409	3.508	0.001**
$R = 0.467$, $R^2 = 0.218$, R^2 adjusted $= 0.147$, $F = 3.060$, Sig = 0.003**					
Family satisfaction					
(constant)	1.583	1.341		1.180	0.243
Gender of the parent/guardian	0.583	0.274	0.287	2.127	0.038
Age of parent/guardian	−0.020	0.022	−0.139	−0.902	0.371
Number of children	0.040	0.171	0.029	0.236	0.815
Gender of the child with cancer	0.330	0.226	0.179	1.459	0.151
Age of child with cancer	−0.006	0.034	−0.032	−0.182	0.856
Children or AYAs	−0.186	0.508	−0.062	−0.365	0.716
Diagnosis time	0.002	0.237	0.001	0.007	0.994
Social support received	0.403	0.204	0.328	10.977	0.050*
Social support provided	0.059	0.185	0.049	0.316	0.753
$R = 0.546$, $R^2 = 0.299$, R^2 adjusted $= 0.175$, $F = 2.411$, Sig = 0.023*					

$^*p \leq 0.05$; $^{**}p \leq 0.01$. 95% CI for the B. Gender: 1 = male and 2 = female; Children or AYA: 1 = 0–14 age and 2 = 15 age and over; Diagnosis time: 1 = 0–2 years and 2 = 3 years and over.

These results confirm the importance of receiving support to cope with daily family tasks as well as facing the different situations derived from the child's disease in a more efficient manner (Choi et al., 2016). However, the increase of life satisfaction is generated by the social support provided by parents to other people, not by the social support received. This might be explained by the fact that on many occasions, those parents who provide support become examples for others (Hombrados-Mendieta et al., 2004). This fact may give parents back some control over their own lives, thus becoming active subjects in the development of resources, facing problematic situations and providing support (Hombrados-Mendieta and Martimportugués, 2006). This phenomenon may directly affect parents' life satisfaction as they may feel they are able to help others thus feeling useful and turning their personal experiences into a potential way of helping themselves and others.

It is important to note that some sociodemographic and clinical variables are key to understand the object of study. There are gender differences in satisfaction with social support received and provided, where male parents feel more satisfied than female parents. This fact is relevant since it could be related to the roles in providing care, that tend to be done by women, who usually play the role of main carers (Cueto et al., 2013). Meaning that they might require higher support, therefore reducing their satisfaction. In this sense, it is also interesting to note that female parents feel higher family satisfaction. Family satisfaction relates to the system, connections and communication that take place between the family members (Sobrino, 2008); female parents might value more family union and communication dynamics that happen within the family. Parents of adolescents and young adults aged 15–21 (AYA) experience less stress than those parents of children aged 0–14.

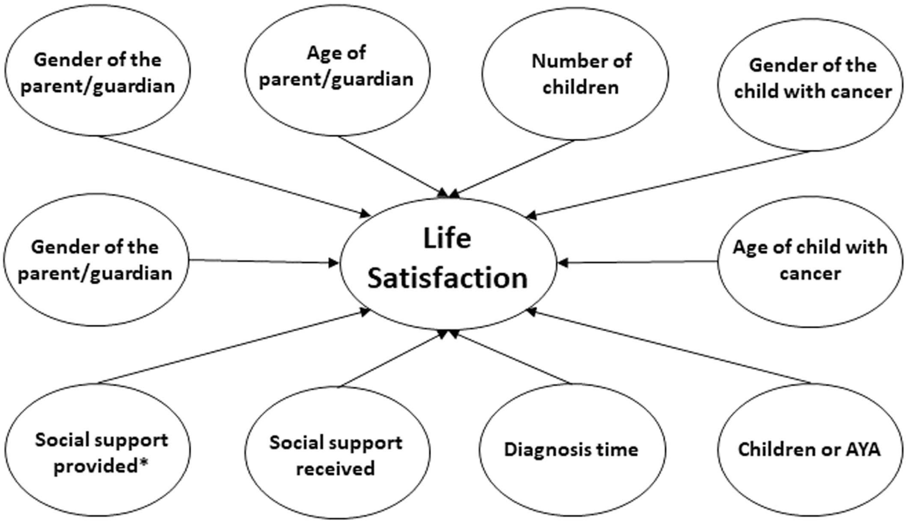

FIGURE 2 | Life Satisfaction model.

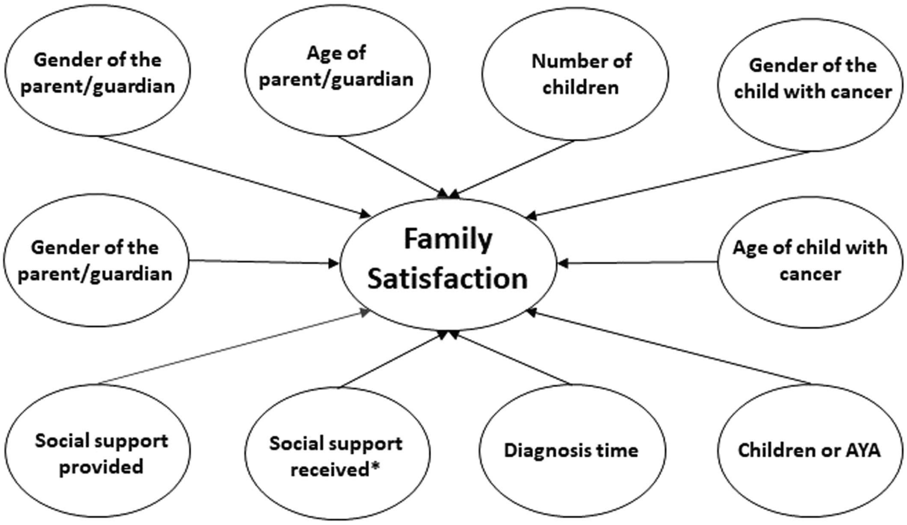

FIGURE 3 | Family Satisfaction model.

This result might be caused by the load of care that an ill child requires, since younger children are more dependent on their parents' care, compared to AYAs, who also require care but are more autonomous in many daily chores. This fact might have relevant implications in parents' perception of stress. It is also of interest to mention that the number of children is a variable that relates with stress perception. This might be due to the fact that having other children in the family means additional sources of support for parents, which might have a positive effect on parents' perception of stress.

We would also like to highlight the fact the results obtained could be of relevant guidance towards achieving a better understanding on the relations between the study's variables, despite the fact that the size of the effect is not very high that the sample studied is very specific, the low incidence in the general population and the fact that studying variables, such as social support provided and life and family satisfaction in parents of children and AYAs with cancer, is considerable new.

Practical Implications

Some guidelines for intervention can be obtained from these findings. Given that support received related to lower levels of stress and higher family satisfaction, intervention guidelines for the family environment could be designed in order to provide parents with the support they need. Individuals usually receive support from their natural networks of support, which are mainly formed by relatives and friends. These natural networks provide a wide range of types of support. However, as noted by Hombrados (2013), sometimes these networks are willing to provide help, but they do not know how. Very often, it is necessary to work with relatives or the closest support network of those who are facing the problematic situation. This is often the case in individuals who suffer from cancer and other severe diseases, as such diseases can alter the support network and feelings of fear or avoidance might appear in the members of support networks. Not knowing which is the appropriate way of taking care of those parents of children with cancer can lead to anxiety. Undesired and opposed effects to the reception of support may give rise to physically avoiding the ill person or avoiding communication about the disease. It is necessary, therefore, to intervene and assess the needs of parents and develop competencies in the provision of help by their closest support network.

Regarding support provided, it has been confirmed that parents feel higher life satisfaction when they provide support to others. Keeping themselves active, helping other parents or participating in associations is likely to make them have active coping mechanisms to face their child's disease. In this sense, intervention guidelines could be designed in order to reduce the helplessness feelings caused by their child's situation through participating and promoting the empowerment of parents.

Sociodemographic and clinical variables of families, the role of other children in the family, the ages of children with cancer (children and AYAs) and the gender of parents should be further explored and studied, since they are tightly linked to the role undertaken by parents as main carers.

Having a broader understanding of the effect of social support received and provided and sociodemographic and clinical variables on stress levels and life and family satisfaction can have highly relevant implications in the design of intervention guidelines that improve parents' life situations. It seems clear that improving parents' satisfaction requires addressing psychosocial aspects (Sánchez-Egea et al., 2019), for which it is necessary to know how these variables relate based on the families that face these situations (Kedia et al., 2020).

Limitations

Among the limitations of the present study, its cross-sectional design is to be noted. For future research, it would be convenient to carry out a longitudinal design in order to better know the relations between variables and how they affect each other. Furthermore, all participants came from Malaga (Spain), which limits the extrapolation of findings to other contexts and cultures. It would be interesting to convey this study in other countries.

Regarding the features of the sample, there were more mothers than fathers, so it would be important to balance the number of mother and father participants to know their needs for support differently.

However, it is important to bear in mind that this fact reflects a social reality—the number of women who take care of their children is significantly higher than the number of men. Moreover, women tend to participate more in this kind of research (García-Calvente et al., 2004).

Other matters to be considered in future lines of research include considering other relevant sociodemographic variables, which might be related to stress management and life and family satisfaction of parents, such as the stage of cancer where children are and the different types of cancer. On the other hand, studying the reciprocity and differences between social support received and provided in future lines of research could also be of interest to find out if such differences are also relevant in coping with cancer in children. Other limitation to consider is the common source bias, which means that the fact that parents could belong to the same family unit in some cases was not controlled. Information on those parents who did not wish to participate was not gathered either, which can also imply a limitation in the generalisation of results.

CONCLUSION

One of the strengths of this study is that novel contributions have been made on the relations between variables related to the process of facing childhood and young adulthood cancer by parents. To broaden the knowledge on these relations, it is key for parents to adapt to the situation caused by the disease, clinical practices and action plans. There are very few quantitative studies that focus on studying the psychosocial variables of parents in the context of childhood and young adulthood psycho-oncology.

It is necessary to consider the practical relevance of the findings from the present study and apply them to the daily tasks of those professionals who provide psychosocial support in these situations. Some of the therapeutic strategies that could be applied by professionals is to know and promote parents' social support networks and know the level of social support they receive and provide. This can help reduce the negative effects of stress and increase levels of life and family satisfaction.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Comité Ético de la Junta de Andalucía. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

IH-M and MM-M: conceptualization and methodology. AM-G and MM-M: formal analysis and writing—original draft preparation. AM-G, IH-M, MM-M, and IR-R: investigation, writing—review and editing, and supervision. IH-M: funding acquisition. All authors have read and agreed to the published version of the manuscript.

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Boys Don't Cry: Examining Sex Disparities in Behavioral Oncology Referral Rates for AYA Cancer Patients

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Psychosocial distress is highly prevalent in cancer patients, approaching rates around 40% across various cancer sites according to multicenter studies. As such, distress screening procedures have been developed and implemented to identify and respond to cancer patients' psychosocial distress and concerns. However, many cancer patients continue to report unmet psychosocial needs suggesting gaps in connecting patients with psychosocial services. Presently, there is a paucity of research examining sex-based disparities in referral rates to behavioral oncology services, particularly for adolescent and young adult (AYA) cancer patients. Informed by gender role conflict and empirical literature documenting disparities in cancer care and treatment based on a variety of sociocultural variables, this study aimed to examine the presence of sex disparities in referral rates to behavioral oncology services for AYA cancer patients. Data for this study consisted of 1,700 AYA cancer patients (age 18–39) who completed a distress screening at a large cancer center of a teaching hospital in the Midwestern United. Results indicated that patient sex significantly predicted the odds of behavioral oncology referral ($\gamma_{50} = -0.95$, Odds ratio $= 2.60$, $p < 0.001$). This finding indicates that female AYA cancer patients are 2.5 times more likely to be referred to behavioral oncology services compared to male AYA cancer patients after controlling for psychosocial distress and emotional, family, and practical problems. Additionally, we found that emotional problems significantly moderated the odds of referral for males and females ($\gamma_{60} = 0.37$, Odds ratio $= 1.44$, $p < 0.001$), however the odds of referral for males who endorsed emotional problems were lower than males who did not endorse emotional problems. This contrasted with female AYA cancer patients where the endorsement of emotional problems increased the odds of referral to behavioral oncology services. Findings are discussed with particular focus on how to enhance equitable care and reduce sex and other sociocultural-based disparities in AYA psychosocial oncology.

Keywords: adolescents and young adults (AYA), access, sex disparities, psychosocial distress, psycho oncology

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INTRODUCTION

Psychosocial distress is highly prevalent in cancer patients, approaching rates between 40 and 60% across various cancer sites according to multicenter studies (Zabora et al., 2001). In a large multicenter study consisting of 55 cancer treatment centers in the United States and Canada, 46% of patients reported significant distress as measured by the Distress Thermometer (Carlson et al., 2019; Essue et al., 2020). In another meta-analysis, prevalence rates of depression, anxiety, and adjustment disorder in cancer patients were 16.5, 9.8, and 15.4%, respectively (Mitchell et al., 2011). Unabated, psychosocial distress in cancer patients has negative sequalae on quality of life, emotional wellbeing, psychosocial functioning, access and adherence to cancer care, and biological outcomes (IOM, 2007; Caruso and Breitbart, 2020). Additionally, unresolved psychosocial problems in cancer patients have adverse economic sequalae on the patient and health care system (Cardoso et al., 2013; Doherty et al., 2019). These are exemplified by longer hospital stays, lack of adherence to scheduled visits and prescribed treatments, increased unplanned visits to emergency rooms with additional imaging and work-up, amongst other avoidable expenses (Carlson and Bultz, 2004; Essue et al., 2020).

These adverse effects prompted national standards of cancer care to include identifying and addressing psychosocial needs of cancer patients as an integral part of cancer care (Jacobsen and Wagner, 2012). Indeed, several agencies, including the American Society of Clinical Oncology, the Canadian Association of Behavioral oncology, and the Institute of Medicine have developed guidelines and recommendations for the implementation of systematic identification of psychosocial distress in cancer patients (Carlson and Bultz, 2003; IOM, 2007). Distress screening processes were developed utilizing validated instruments and tools to identify and assess the severity of psychological distress in cancer patients coupled with recommendations for triage processes to ensure interventions and follow up are implemented (Loscalzo et al., 2013; Donovan et al., 2014; Bultz, 2017). Despite these advancements in the practice of identifying and treating psychosocial distress, important limitations exist (Carlson, 2013; Bultz et al., 2021) and many cancer patients continue to report unmet psychosocial needs (McMurtry and Bultz, 2005; Meggiolaro et al., 2021). This is partly attributable to ineffective psychosocial distress screening procedures, patients' help-seeking behaviors, limited access or availability of psychosocial services, as well as provider attitude and cultural factors (Dilworth et al., 2014; Brebach et al., 2016; Carolan et al., 2018). The discrepancy between heightened psychosocial needs and the bridging of patients with psychosocial services is especially pronounced in some minority groups and vulnerable populations who bear the brunt of psychosocial adversity collateral to the physical impact of a diagnosis cancer (Kamen et al., 2017; Kent et al., 2019).

One understudied barrier to behavioral oncology care is gender/sex bias and disparities. A large body of research exists documenting disparities in oncology treatment and care based on various sociocultural identities, such as race (Emerson et al., 2020; Hardy and Du, 2021), socioeconomic status (SES) (Dreyer et al., 2018; Karanth et al., 2019), and gender (Tabaac et al., 2018; Benchetrit et al., 2019) however, less attention has been paid to sociocultural disparities, namely gender/sex disparities, in referral practices to behavioral oncology services. Given previous research documenting disparities in cancer care, and the need to increase utilization rates of behavioral oncology for cancer patients with psychosocial distress, this study aims to examine the presence of sex disparities in referral rates to psychosocial services (i.e., counseling and psychiatric services) for Adolescent and Young Adult (AYA) cancer patients.

AYA Psychosocial Needs

AYA cancer patients are recognized as an underserved minority within cancer patients whose unique developmental and social attributes put them at a particular disadvantage related to social, interpersonal, academic, occupational, and financial sequalae of a cancer diagnosis further deepening the chasm in cancer care between them and their non-AYA counterparts (Clinton-McHarg et al., 2010; Jacobs et al., 2018).

Longitudinal data demonstrates patterns of disproportionate and lasting financial burden, consequences of interrupted education and work, difficulties with relationships and family planning, and unresolved physical and mental health issues related to AYA history of cancer (Smith et al., 2019). Evidence also suggests that the mere availability of psychosocial services for AYA patients is not a factor in reducing and preventing future psychosocial dysfunction in this patient population (Patterson et al., 2017; Jacobs et al., 2018) calling for a higher level of communication and triage of these patients to psychosocial services. Ongoing efforts include thoughtful evaluation of age-specific screening tools for identifying psychosocial distress in AYA, strategies honing in on areas of success and where improvement is needed to produce interventions that are specifically tailored to this age group.

Barriers to Behavioral Oncology

Despite effective screening tools and the proliferation of evidence-based care for cancer patients, problems remain in the equity of service delivery and referral processes. Psychosocial oncology literature has demonstrated that disparities exist in psychological distress screening, referral of patients to psychosocial resources, and the utilization of such resources by patients (Kamen et al., 2017; Nolan et al., 2018). Barriers to access psychosocial services are multifaceted and include systemic, patient, and provider factors, such as low referral rates by physicians, patients' perceived stigma about accessing mental health services, and fragmentation of care (Matthews et al., 2004). For example, Dilworth et al. (2014) conducted a systematic review and found that physicians' negative perceptions about psychosocial services were one of the most common barriers to care. These negative perceptions stemmed from the perceived lack of evidence-backed research for psychosocial interventions, the potential to cause psychosocial harm, and the priority to control cancer-related symptoms over psychosocial care (Dilworth et al., 2014). In another study, patient age was found to significantly contribute to the likelihood of referral to psychosocial care in a sample of metastatic cancer patients, such that younger patients were more likely to be referred to psychosocial services (Ellis et al., 2009). Regarding utilization rates of psychosocial services, one study found that the majority of cancer patients attending counseling services were well-educated, urban-residing women (Nekolaichuk et al., 2011). Together, this body of research suggests that some cancer patients may be more or less likely to be referred to psychosocial services based on demographic factors, such as age, education, and SES. Regarding research that has found that on average, women cancer patients utilize psychosocial services at greater rates compared to men, it may be that women are more likely referred to these services compared to men resulting in differential utilization rates. Gender role conflict and social constraint may explain this phenomenon, and support research examining the presence of gender disparities in behavioral oncology referral rates (Strong et al., 2007; Salk et al., 2017).

Gender Role Conflict

Gender role conflict (GRC) and traditional masculinity norms are important factors that can lead to compromised adjustment in men with cancer (Nicholas, 2000). As noted by O'Neil et al. GRC refers to the negative cognitive, emotional, and behavioral consequences associated with male socialization (O'Neil et al., 1986). Specifically, O'Neil defined GRC as "a psychological state in which gender roles have negative consequences or impact on the individual or on others" (p. 25) (O'Neil, 1990). Within a cancer diagnosis context, research has demonstrated how traditional and restrictive masculinity and gender role conflict are related to poorer physical and psychological outcomes in men with cancer (Maliski et al., 2008; Hoyt, 2009). In one study, cancer-related masculine threat was associated with poorer physical outcomes over time in a sample of men with prostate cancer (Hoyt et al., 2013).

One theorized mechanism through which gender role conflict may negatively influence physical and psychological outcomes of men with cancer is emotional approach coping (Lennon et al., 2018). Emotional approach coping has been defined as identifying, understanding, and expressing emotions appropriately and is posited to consist of two strategies, emotional processing, and emotional expression (Lennon et al., 2018). Interestingly, Hoyt et al. (2013) also found that cancer-related masculine threat was significantly associated with decreased emotional processing, which ultimately explained the effect of cancer-related masculine threat on poor physical outcomes. In another study, gender role conflict was found to significantly predict distress in a sample of men with prostate cancer (Lennon et al., 2018). Together, these findings suggest that gender role conflict and emotional approach coping, or the tendency for men to restrict emotional expression, may contribute to negative cancer-related physical and psychological outcomes. Simultaneously, gender role conflict may also impact the likelihood that providers and care team members will refer men to behavioral oncology services as talking about emotional distress with men may violate traditional male gender role socialization (Vogel et al., 2014).

To our knowledge, no study has examined the presences of sex disparities in referral rates to behavioral oncology services, nor has this question been studied in the AYA population. This is important given literature suggesting that younger adult males tend to experience more gender role conflict than older adult males (Watts and Borders, 2005). One study found that young adult men who reported greater restrictive emotionality endorsed lower levels of resiliency in the face of adverse experiences (Galligan et al., 2010). In another study, Pederson and Vogel (2007) examined several mediators of the relationship between gender role conflict and college-aged men's willingness to seek counseling. Results indicated that men who experienced greater gender role conflict were less likely to disclose distressing information, which subsequently led to less positive attitudes and willingness to seek counseling. These studies are important as they highlight the fact that gender role conflict occurs across the lifespan and may have unique consequences particularly for AYA men.

Purpose of Study

This study examines the presence of sex-based disparities in referrals to behavioral oncology within an AYA cancer patient population. Research has documented the importance of screening and responding to cancer patients' psychosocial distress. However, research has demonstrated how distress screening and referral efforts may not be equitable across all cancer patients. Yet, few studies have examined sociocultural barriers to access for AYA cancer patients. Particularly, there is a paucity of research examining if referral rates to behavioral oncology services vary based on patients' sex in AYA cancer patient populations. Informed by gender role conflict and empirical literature documenting disparities in cancer are and treatment based on a variety of patient demographic variables, this study aimed to examine the presence of sex disparities in referral rates to behavioral oncology services for AYA cancer patients, including individual counseling or psychiatric services. It is important to note that sex and gender represent distinct constructs and should not be used interchangeably. Gender is the range of characteristics pertaining to, and differentiating between femininity and masculinity, whereas sex refers to an individual's biological makeup resulting in a male or female phenotype. While the spectrum of gender identities (and even biological sex) is wide, this study focuses specifically on sex disparities (i.e., male and female) due to the data available in our archival data set which was limited to the patients' reported sex, rather than their gender identity. However, despite this limitation, we believe that examining sex-based disparities informed by gender role conflict is an important endeavor in the field of psychosocial oncology. Given the aforementioned literature on the role of gender role conflict on cancer related outcomes for men, we proposed the following hypothesis.

Hypothesis 1: Sex disparities will exist in behavioral oncology referrals, such that male AYA cancer patients will have lower odds of being referred to behavioral oncology services compared to their female AYA cancer patient counterparts.

As a second aim, we sought to examine male AYA cancer patients' endorsement of emotional problems on the likelihood that they would be referred to behavioral oncology services. Emotional approach coping is theorized to serve as an important factor in male cancer patients' health outcomes, as well as their willingness to seek treatment. It may be that male AYA cancer patients are less likely to be referred to behavioral oncology services because they are less likely to disclose distressing information as a result of emotional approach coping (Pederson and Vogel, 2007). Informed by the theory of emotional approach coping we proposed the following hypothesis.

Hypothesis 2: The odds of male AYA cancer patients being referred to behavioral oncology care will significantly vary as a function of their endorsement of emotional problems, wherein male AYA cancer patients who endorse emotional problems will be more likely to be referred to services compared to male AYA patients that do not endorse emotional problems.

METHODS

Participants

The data for this study consisted of 1,700 AYA patients (age 18–39) with a diagnosis of cancer at a large cancer center of a teaching hospital in the Midwestern United States. Patient characteristics are reported in **Table 1**. The average age was 30.38 (SD = 6.08) with a range of 18–39 years old. 70.4% of the sample were female (n = 1,197) and 29.6% were male (n = 503). Regarding race/ethnicity, 83% (n = 1,411) patients identified as white, 8.2% (n = 139) identified as Black/African American 2.8% (n = 48) identified as Hispanic/Latino/a, 2.7% (n = 46) identified as Asian/Asian American, 1.4% (n = 24) identified as Multiracial, <1% (n = 6) American Indian/Alaska Native, <1% (n = 2) identified as Native Hawaiian/Pacific Islander, and 1.5% (n = 24) did not report their race/ethnicity. Regarding oncology department, 1,076 (63.3%) patients were seen in hematology oncology, 82 (4.8%) in pulmonary oncology, 348 (20.5%) in gynecology oncology, 149 (8.8%) in surgical oncology, and 45 (2.6%) in urology oncology. The 1,700 patients were seen by 51 nurses. No demographic data for oncology providers or nurses was available in the data set.

TABLE 1 | Patient characteristics.

Characteristics	***n* (%)**
Age, mean (SD)	30.38 (6.08)
Male	503 (29.6)
Female	1,197 (70.4)
Race/ethnicity	–
White	1,411 (83.0)
Black/African American	139 (8.2)
Hispanic/Latino/a	48 (2.8)
Asian/Asian American	46 (2.7)
Multiracial	24 (1.4)
American Indian/Alaska Native	6 (<1.0)
Native Hawaiian/Pacific Islander	2 (<1.0)
Department	–
Hematology Oncology	1,076 (63.3)
Pulmonary Oncology	82 (4.8)
Gynecology Oncology	348 (20.5)
Surgical Oncology	149 (8.8)
Urology Oncology	45 (2.6)

Measures

Distress Screening Questionnaire

The distress screening questionnaire is an adapted version of the NCCN distress thermometer (DT) which consists of the single question rating the level of distress of patients over the week prior to their visit, associated with a problem list requiring yes/no answers to the presence of difficulties in three domains: practical problems (i.e., financial, transportation, insurance, etc.), family problems (i.e., problems with partner, siblings), and emotional problems (i.e., depressive, anxiety, sleep problems as well as existential questions and ambivalence about spirituality). The distress thermometer is a one item visual analog that assesses individual's distress level from 1 to 10, where 1 is low distress and 10 is high distress.

Behavioral Oncology Referral

Referral rates to behavioral oncology services were accessed through patient medical records. Specifically, a medical record review allowed the research team to identify which patients had an order placed for behavioral oncology and which patients did not (i.e., referral vs. no referral). Behavioral oncology services consisted of either individual counseling or psychiatric care. Referral to behavioral oncology services was dummy coded (0 = no referral, 1 = referral).

Procedures

At the participating cancer center all new cancer patients are assigned a distress screening questionnaire prior to their first appointment, and every 3 months upon follow up, to assess psychosocial distress and concerns. The distress screening questionnaire consists of the distress thermometer and three yes/no questions assessing the presence of practical problems (i.e., financial, transportation, insurance, etc.), family problems, and emotional problems. Archival data for this study was accessed through the electronic medical record system of the participating site. Archival data included patient demographic data, appointment data, distress screening data, including patient distress scores and problems indicated and order status for behavioral oncology (i.e., referral placed). Behavioral oncology services consisted of individual counseling services with a licensed mental health provider or psychiatric services with a psychiatry provider. All procedures were approved by the first author's institutional review board.

Data Analysis Plan

Data was analyzed using multilevel logistic modeling to account for the nested nature of our data (i.e., patients nested within nurses) and examine the odds of referral to behavioral oncology services for male identified and female identified AYA cancer patients. Specifically, we used Hierarchal Linear Modeling (HLM; Raudenbush et al., 2011) to run a 2-level model with patient sex (0 = male, 1 = female) as a level-1 predictor of referral to behavioral oncology (0 = no referral, 1 = referral). Patients' distress thermometer score, and presence of emotional, family, and practical problems (0 = no, 1= yes) were entered as covariates at level 1. In addition to modeling fixed effects of the overall odds of referral based on patients' sex, we examined the variability in the odds of referral based on patient sex between nurses at level 2. Specifically, we included a random component at level 2 to examine between-nurse variability in the odds of male and female AYA cancer patients being referred to behavioral oncology. To test our second hypothesis, we included an interaction term between emotional problems and patient sex in our model. This interaction term tested whether the odds of being referred to behavioral oncology varied as a function of male AYA cancer patients' endorsement of emotional problems.

RESULTS

Overall, the average distress score was 3.38 (SD = 2.82), with an average score of 2.90 (SD = 2.74) for males and 3.59 (SD = 2.84) for females. On average, 40.0% of patients endorsed practical problems, 18% endorsed family problems, and 45% endorsed emotional problems, with 41, 12, and 38% of males endorsing practical, family, and emotional problems, respectively, and 40, 20, and 48% of females endorsing practical, family, and emotional problems, respectively.

Results from the multilevel logistic analysis of patient sex on the odds of behavioral oncology referral are reported in **Table 2**. Our first hypothesis that—male AYA cancer patients will have lower odds of being referred to behavioral oncology services compared to their female AYA cancer patient counterparts—was supported. Specifically, patient sex significantly predicted the odds of behavioral oncology referral ($\gamma_{50} = 0.95$, Odds ratio = 2.60, $p < 0.001$). This finding indicates that female AYA cancer patients were ∼2.5 times more likely to be referred to behavioral oncology services compared to male AYA cancer patients after controlling for distress levels and emotional, family, and practical problems. All random components at level 2 were not significant.

Our second hypothesis that—the odds of male AYA cancer patients being referred to behavioral oncology care will significantly vary as a function of their endorsement of emotional problems, wherein male AYA cancer patients who endorse emotional problems will be more likely to be referred to services compared to male AYA patients that do not endorse emotional problems—was supported, but in the opposite direction. Specifically, we found that emotional problems significantly moderated the odds of referral for males and females ($\gamma_{60} = 0.37$, Odds ratio = 1.44, $p < 0.001$), however the odds of referral for males who endorsed emotional problems were lower than males who did not endorse emotional problems. As seen in **Figure 1**, this contrasts with female identified AYA cancer patients where the endorsement of emotional problems increased the odds of referral to behavioral oncology services.

DISCUSSION

This is one of the first studies to examine the presence of sex disparities in access to behavioral oncology services for AYA cancer patients. As hypothesized, our results indicated that sex disparities in behavioral oncology referral rates exist for AYA cancer patients, such that female AYA cancer patients have significantly higher odds of referral to behavioral oncology compared to their male AYA counterparts. This study is meaningful as it adds to the limited literature on sociocultural disparities in behavioral oncology for AYA cancer patients who are disproportionately impacted by adverse psychosocial sequalae of cancer (Nass et al., 2015; Smith et al., 2019). These findings support previous research that has documented disparities in psychosocial care and inequity of service delivery and referral processes to behavioral oncology services. As referenced earlier, a burgeoning body of research has documented unequitable practices in distress screening, referral of patients to psychosocial resources, and the utilization of such resources by patients (Ellis et al., 2009; Nekolaichuk et al., 2011; Kamen et al., 2017; Nolan et al., 2018).

Interestingly, our second finding indicated that endorsing emotional problems on the distress screening increased the odds of referral to behavioral oncology for female AYA patients but decreased the odds of referral for male AYA patients. Although we were unable to disentangle provider and patient factors that may contribute to sex disparities in accessing behavioral oncology services, this finding suggests that even when male AYA patients do not endorse traditional gender roles and disclose emotional problems, they are less likely to be referred to behavioral oncology care. It may be that despite male AYA patients' ability to disclose emotional distress, providers overlook these concerns related to their own gender role socialization. Acknowledging and discussing emotional concerns with men is often proscribed in the US, and therefore, providers acknowledging and further assessing male AYA cancer patients' emotional concerns would go against these gendered social norms. However, it is important to note that this explanation of our findings is post hoc, and future research is needed to understand this finding and possible explanation further.

Access to psychosocial services is influenced by system, provider, and patient factors (Matthews et al., 2004) and a large body of research has documented disparities in oncology treatment and care based on various sociocultural identities, such as race (Emerson et al., 2020; Hardy and Du, 2021), SES (Dreyer et al., 2018; Karanth et al., 2019), and gender (Tabaac et al., 2018; Benchetrit et al., 2019). Access may also be subject to provider biases based on attitudes, socialized norms, as well as patients' perceived stigma around mental health services and varying communication styles and willingness to disclose emotional and psychosocial distress.

Implications for Practice and Future Research

There are several implications for AYA behavioral oncology care given the findings of this study. First, disparities in referral rates between male and female AYA patients maybe

TABLE 2 | Multilevel logistics model of sex disparities in behavioral oncology referrals.

Variable	Estimate	*SE*	Odds ratio	*df*	*p*-value
Behavioral oncology referral, γ_{00}	−3.05	0.06	0.05	50	< 0.001
Distress thermometer, γ_{10}	0.02	0.01	1.02	50	0.060
Practical problems, γ_{20}	−0.11	0.03	0.90	50	0.003
Family problems, γ_{30}	0.17	0.05	1.19	50	< 0.001
Emotional problems, γ_{40}	−0.04	0.03	0.96	50	0.175
Patient sex, γ_{50}	0.95	0.07	2.60	50	< 0.001

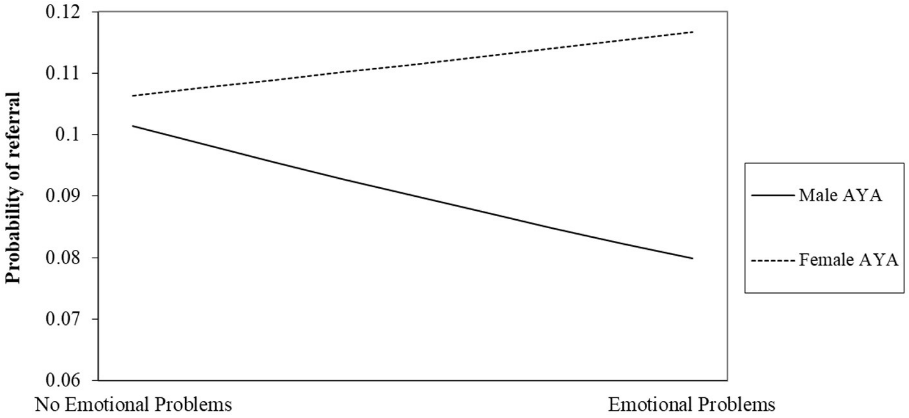

FIGURE 1 | Interaction between emotional problems and sex on odds of referral to behavioral oncology.

partially related to gender role conflict, which posits men are socialized to avoid emotions and behaviors that are considered to be less masculine. These behaviors may include discussing one's emotional symptoms or outwardly expressing one's inner emotional state. As such, it may prove beneficial to intervene with male AYA cancer patients to examine and address socialized gendered behaviors and attitudes about help-seeking to increase their willingness to disclose distressing experiencing with their care team and seek services when available. This may include psychoeducation for AYA cancer patients on the prevalence of psychosocial concerns within this population, the availability of behavioral oncology services, and the effectiveness of these services. Additional interventions can include narrative based health messaging that highlights the experiences of other male AYA cancer patients disclosing their emotional concerns and their experiences of seeking help.

A second factor that may explain sex disparities in access to behavioral oncology is provider attitudes and socialized gender roles. As discussed above, providers and other oncology staff may be less likely to offer a referral to behavioral oncology services if providers themselves have been socialized to avoid discussing emotions related to cancer with their male patients for fear of undermining masculinity (Vogel et al., 2014). As such, interventions aimed at exploring providers socialized gendered expectations and attitudes toward mental health and help seeking may be effective to increase awareness of potential biases. These may include provider education about the cost of unresolved psychosocial adversity in males and females alike, and the impact of dismissing male psychosocial distress and the prevalence of working-age men with cancer who go without having their psychiatric problems treated due to a variety of factors (Akechi et al., 2020). These interventions may help to reduce provider bias and increase their willingness to acknowledge, explore, and validate emotional concerns and refer to appropriate behavioral oncology services for all AYA cancer patients, including men.

Additional studies examining patient and provider factors of gender/sex disparities in access to behavioral oncology care are needed. Specifically, studies examining patient and provider attitudes about mental health and help-seeking may prove helpful for enhancing our understanding of critical factors to address in increasing access to services for all AYA cancer patients. Tools such as the Gender Role Conflict Scale (GRCS) and its short form could be used to examine both provider and patient gender role conflict and their willingness to disclose and explore psychosocial functioning and distress as well as the likelihood of referral and treatment seeking (O'Neil et al., 1986; O'Neil, 2015). Related, studies can examine the interactions/communication between patients and providers that may contribute to help-seeking. Advances in machine learning and natural language processing technologies may make this possible and could assist in identifying important factors within the patient-provider interaction that could increase access to behavioral oncology care. Lastly, research will need to ultimately test the effectiveness of interventions aimed at increasing access to behavioral oncology services, especially for male AYA cancer patients. Beyond identifying factors contributing to gendered disparities in accessing behavioral oncology care, research is needed to determine if these factors are malleable and if which interventions are effective in addressing these disparities and increasing access.

Limitations

With any study, there are limitations. One limitation of this study is the use of a single site to examine disparities in access to behavioral oncology care. Although we used a relatively large sample, our findings may be related to site-specific phenomenon and factors and may not generalize to other cancer centers and systems. Future research can address this by replicating these findings with a multi-center design and larger sample size. Another limitation is that our study used archival data obtained from an electronic medical record (EMR) database and are therefore subject to data quality and accuracy issues often present when using EMR data for research. Additionally, sex was obtained from the EMR and may be limited in comparison to patient's gender. For example, patients' gender identity and expression may differ from their sex assigned at birth and sex was defined as a binary construct (Freiburger, 2018). We believe this not only represents an important limitation of our study, but the larger healthcare system as a whole. Moving forward, healthcare and EMR systems could allow patients to record their sex and gender outside of a binary construct as an affirming practice for patients with diverse gender identities. Related, our findings are only generalizable to male and female AYA patients, and future research is needed to explore these findings regarding gendered disparities to psychosocial services, particularly for gender minority patients. Another limitation is that we do not know what occurred in the appointments with patients and providers, which is important data that may help to further explain the presence of sex disparities. Finally, referrals to behavioral oncology services were recorded as a consultation order for behavioral oncology within the EMR, which would not have captured patients who were referred to outside providers, or those who were already receiving care for psychosocial needs through services outside of our site (i.e., community-based mental health centers).

CONCLUSION

Disparities in psychosocial services for AYA cancer patients mirror inequitable services seen in other disciplines of medicine and have pervasive consequences for patients. These disparities can stem from systemic, provider and patient factors, and can further exacerbate poor psychosocial and health outcomes. Interventions highlighting the value and availability of psychosocial services to AYA patients, minimizing stigma around mental health, and addressing unconscious bias is imperative to foster equity in the access to psychosocial services. It is our hope that this study sheds light on the prevalence of gendered disparities in access to behavioral oncology for male AYA cancer patients and encourages future research to address inequities in access to care.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by University of Iowa Internal Review Board. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

AA provided oversight, supervision of research project and team, conceptualized the study, managed data, wrote manuscript, and prepared for submission. MK conceptualized study, analyzed data, wrote manuscript, and prepared for submission. JB wrote manuscript and prepared for submission. All authors contributed to the article and approved the submitted version.

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The Clinical Utility of the Adolescent and Young Adult Psycho-Oncology Screening Tool (AYA-POST): Perspectives of AYA Cancer Patients and Healthcare Professionals

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Objective: Routine psychosocial screening and assessment of people diagnosed with cancer are crucial to the timely detection of distress and provision of tailored supportive care; however, appropriate screening tools have been lacking for adolescents and young adults (AYAs), who have unique needs and experiences. One exception is the recently validated AYA Psycho-Oncology Screening Tool (AYA-POST) for use with young people aged 15–29 years, which comprises a distress thermometer and age-specific needs assessment. This study investigates the clinical utility of this measure, as well as the subsequent service responsiveness within the Australian Youth Cancer Services.

Method: In total, 118 AYAs and 29 healthcare professionals: (HCPs) completed surveys about the clinical utility of the AYA-POST; a subset of 30 AYAs completed a 3-month follow-up survey assessing service responsiveness. Descriptive statistics (frequencies/means) were computed for all items, with chi-square analyses used to explore whether perceived clinical utility varied with AYA age, AYA sex, HCP discipline or HCP length of time using the AYA-POST.

Results: Participants' responses demonstrate high levels of satisfaction with the tool, evidencing its appropriateness, practicability and acceptability. Moreover, the AYA-POST was reported to facilitate communication about psychosocial needs and prompt referrals, indicating good service responsiveness. Ratings of clinical utility did not differ significantly between AYA and HCP groups.

Conclusion: This study demonstrates that the AYA-POST is an appropriate tool in the psychosocial screening of AYAs with cancer, facilitating the identification of distress and unique concerns in this population and valuable in triaging and tailoring care for young cancer patients.

Keywords: adolescent and young adult, clinical utility, distress, needs assessment, psycho-oncology, psychosocial screening

INTRODUCTION

A cancer diagnosis in adolescence or young adulthood can cause significant psychosocial disruption during an already dynamic developmental stage. Affected adolescents and young adults (AYAs, 12–25 years) are at greater risk of developing mental health conditions (Barnett et al., 2016; Zebrack et al., 2016), experience disruptions to familial, peer and romantic relationships (Warner et al., 2016) and may have their educational and vocational plans interrupted (Fardell et al., 2017). AYAs with cancer typically report higher levels of psychological symptomatology than other age groups (Li and Deng, 2004; Cardoso et al., 2012; Barnett et al., 2016) and the risk of poorer psychosocial outcomes is particularly pronounced for females, AYAs with poorer physical health or late effects, and those experiencing educational/work disruption or financial precarity (Phillips-Salimi and Andrykowski, 2013; Sansom-Daly and Wakefield, 2013; Yanez et al., 2013). Importantly, distress has been linked to lower health-related quality of life (Greup et al., 2018), greater stress (Hodgson et al., 2021), poorer coping and resilience (Xie et al., 2017; Greup et al., 2018; Hodgson et al., 2021) and lower treatment adherence (Robertson et al., 2015). This is particularly concerning as 50–95% of AYAs reportedly experience unmet supportive care needs (Keegan et al., 2012; Xie et al., 2017) which may persist for years beyond the completion of active treatment (Millar et al., 2010) contributing to ongoing distress (Zebrack et al., 2014). It is therefore crucial to identify and address distress and other contributing and compounding psychosocial issues in AYAs with cancer, to minimise negative impacts and facilitate adjustment and wellbeing.

Routine screening and assessment of the psychosocial wellbeing of AYAs with cancer are crucial to the provision of quality, tailored supportive care and guides responsive and efficient service delivery (Palmer et al., 2014; Zebrack et al., 2016; Patterson et al., 2018; Osborn et al., 2019). In particular, psychosocial screening of all patients can help healthcare professionals (HCPs) to identify those experiencing distress and other concerns in a timely and proactive manner, allowing early intervention to address these issues (Butow et al., 2015; Patterson et al., 2018; Riba et al., 2019). However, the implementation of effective screening, assessment and care pathways relies on the availability of robust, validated psychometric measures to detect distress, something which has historically been lacking for AYAs with cancer (Clinton-McHarg et al., 2010; Wakefield et al., 2013). While there has been some examination of the use of adult distress measures with AYA participants (e.g. Chan et al., 2018), until recently no age-specific tools had been validated across the full AYA age range (Wakefield et al., 2013; Patterson et al., 2021a). When selecting appropriate measures of distress for AYAs with cancer, it is crucial that they reflect the unique needs and experiences of the population, in addition to being psychometrically robust and sensitive to change (Clinton-McHarg et al., 2010).

The AYA Psycho-Oncology Screening Tool

In 2008, the Australian National Service Delivery Framework for AYAs with Cancer identified the development of age-specific psychosocial assessment tools and processes as a key priority for care (Australian Government, Cancer Australia, and CanTeen, 2008), leading to the subsequent development of the AYA Oncology Psychosocial Care Manual (Canteen, 2011) which includes the AYA Psycho-Oncology Screening Tool (AYA-POST; Palmer et al., 2014; Patterson et al., 2021b; see **Supplementary Table 1**). This validated tool for young people aged 15–29 years comprises the Distress Thermometer (DT; a single-item measure of psychological distress) and the Needs Assessment (NA; asking patients to indicate if they are concerned about commonly reported concerns). The DT is identical to that used with adult cancer patients: it is recommended for use by the (US) National Comprehensive Cancer Network, has been translated into over twenty languages, with extensive validation work evidencing its strong psychometric properties with adults including sensitivity, specificity and predictive value (Carlson et al., 2012; Donovan et al., 2014). Typically, a cut-off score of 4 on the DT has been used to indicate clinically significant levels of distress in adults (Jacobsen et al., 2005; Donovan et al., 2014), while a cut-off of 5 is more appropriate for AYAs (Patterson et al., 2021b). The accompanying NA was adapted from the adult Problem Checklist (PCL) following indications that the latter did not reflect key AYA concerns (Palmer et al., 2014). Young people with cancer and AYA healthcare professionals consulted on the revision of the original checklist which resulted in fifty issues pertinent to this population spanning six domains: practical needs, family, emotions, social issues, physical symptoms and information (Palmer et al., 2014; Patterson et al., 2021b). The tool also includes an option for AYAs to specify additional concerns they experienced which are not covered on the list. The AYA-POST also includes a checklist of 11 items for clinicians to indicate whether they have discussed key issues with the AYA patient (e.g. clinical trials and fertility preservation), and a joint sign-off by the clinician and AYA to confirm they have completed the tool, understand the process and have been informed of next steps (Patterson et al., 2018).

The AYA Oncology Psychosocial Care Manual and AYA-POST are used nationally in Australia by the hospital-based Youth

Cancer Services (Patterson et al., 2021a) and have been recommended for use by the Clinical Oncology Society of Australia (Psychosocial Management of AYA Cancer Patients Working Group, 2011), as well as being translated for use internationally. The AYA-POST has recently been validated with an international cohort of AYAs with cancer, where it was found to have good convergent validity, with a DT cut-off score of 5 providing acceptable specificity and sensitivity scores for use as a screening tool, and the NA items being highly relevant to this age group (Patterson et al., 2015, 2021b). While this is important and necessary, it is not sufficient in ensuring a tool is useful for clinical practice; its clinical utility also needs to be examined.

Clinical Utility

Smart (2006) conceptualises clinical utility as 'a multidimensional judgement about the usefulness, benefits and drawbacks of an intervention', identifying four key components: appropriateness, accessibility, practicability and acceptability (Smart, 2006). In brief, appropriateness comprises both evidence of efficacy of an instrument and perceptions of its relevance to a particular population. Accessibility covers both economic and logistical issues around resourcing—the procurement and cost of materials. Practicability assesses the functionality and suitability of the materials, as well as if users have the knowledge or training to use them. Finally, acceptability is assessed from the perspectives of clients, HCPs and broader society. In addition, it is also important to consider service responsiveness. The efficacy of distress screening programmes depends not only on the use of screening to identify patients in need, but also further assessment of psychosocial issues, triaging to appropriate services and evidence-based treatment (Carlson, 2013), which is 'where the real impact [of screening] is felt' (Smith et al., 2018). Notably, service responsiveness is context-specific and does not meaningfully generalise beyond the service or programme which is evaluated.

Research into the clinical utility of the DT and PCL in adult populations has thus far focused on its ability to accurately identify patients experiencing clinically significant levels of distress and, to a lesser extent, its acceptability; relatively little work has explored its accessibility, practicability or service responsiveness (Snowden et al., 2011). It is unclear whether use of the DT/PCL improves patient outcomes, perhaps because screening has not consistently led to increased referrals for support in these implementation studies (Snowden et al., 2011). More recent research has largely replicated these findings (e.g. Hollingworth et al., 2013; Williams et al., 2015; Linehan et al., 2017; Van der Meulen et al., 2018), confirming the acceptability of the measures to patients and HCPs but drawing further attention to the need to consider service responsiveness in tandem with more commonly investigated aspects of clinical utility, as the benefits of screening are contingent on its use to provide referrals to appropriate support services and facilitate their uptake.

Since the experience of completing a questionnaire or participating in clinical research may differ based on participant characteristics such as age and gender (Lee et al., 2013; Knäuper et al., 2016), and/or features of the researcher/administrator such as qualifications or expertise (Kost et al., 2011), exploring individual differences relating to these factors, have the potential to provide useful additional information about the generalizability of clinical utility findings.

Present Study

This study is the first to explore the clinical utility of the AYA-POST and subsequent service responsiveness of the Australian Youth Cancer Services (YCS). Perspectives were sought from both AYAs receiving care within the YCS and the HCPs who work with them. The primary aim is to evaluate the appropriateness, practicability and acceptability of the AYA-POST, as well as the service responsiveness of the YCS; a secondary aim was to explore whether perceptions of clinical utility varied between subgroups of AYAs (by gender or age) and HCPs (by discipline or length of time using the AYA-POST). The data collected in this study are part of a larger study that examined the validity of the AYA-POST and identified predictors of distress and psychosocial concerns (Patterson et al., 2015).

MATERIALS AND METHODS

Setting

In Australia, healthcare is provided by a combination of public and private health systems: citizens and permanent residents are able to access universal healthcare through Medicare, which allows free or subsidised access to medical services, hospital treatment and prescription medications (Australian Government Department of Health, 2020), while private health insurance allows greater choice of practitioners and hospitals, and covers other health services and expenses (e.g. physiotherapy and psychology) (Australian Government, 2019). These systems are supplemented by non-government organisations providing health information, counselling services and peer support, among other services (e.g. Lifeline for crisis support and suicide prevention; state Cancer Councils for cancer information and support). The majority of AYAs with cancer are treated in public hospitals (Osborn et al., 2013). Approximately 75% of those requiring hospital-based care are treated through the specialised Youth Cancer Services (YCS), which provide age-appropriate, holistic cancer care to 15–25 year olds across Australia (CanTeen Australia, 2015, 2017; Patterson et al., 2021a). The YCS comprises five jurisdictions covering all Australian states and territories, which have lead sites in major hospitals and work in collaboration with a network of hospitals, health services and HCPs around the nation, allowing AYAs to benefit from both the age-specialised care offered by the YCS and disease-specific expertise of local cancer teams (Patterson et al., 2021a). A key feature of the YCS is their integrated, multidisciplinary approach to cancer care, with teams comprising medical, nursing, allied health and support professionals, and close ties with community organisations (e.g. Canteen for AYA-specific cancer information and support; Patterson et al., 2021a). The psychosocial care pathway implemented in the YCS includes routine screening, assessment and care planning, as detailed in the AYA Oncology Psychosocial Care Manual (Canteen, 2011), to ensure that the concerns of AYAs are detected and addressed in a timely and systematic way (Patterson et al., 2018, 2021a). The use of the AYA-POST is recommended as part of the screening process. Critically, the interconnected and multidisciplinary nature of the YCS provide a rich environment for YCS patients' psychosocial needs to be identified and addressed through internal and external referrals, optimising service responsiveness.

Design

The clinical utility of the DT was evaluated in accordance with Smart's multidimensional clinical utility framework, incorporating both AYA and HCP perspectives.

AYA data collection involved surveys at two time points. The T1 survey was completed within 3 months of diagnosis and included demographic and cancer details, the AYA-POST and questions on the clinical utility of the measure, adapted from Breen et al.'s (2012) work. This survey also contained measures used in the broader validation study (see Patterson et al., 2015, 2021b for further details). The T2 survey was an optional component of the study, intended to be completed by a subset of T1 participants during a follow-up phone interview approximately 2 months later. This survey included the re-administration of the AYA-POST as well as questions on service responsiveness.

HCP perspectives were collected using an online survey, which included questions about the clinical utility of the AYA-POST and barriers to screening for distress.

The study received ethical approval from the Human Research Ethics Committees at seven lead sites across the country: ACT Health (ETH.11.14.331), Children's Health Queensland Hospital and Health Service (HREC/14/QRCH/374), Northern Territory Department of Health and Menzies School of Health Research (HREC-2014-2,275), Peter MacCallum Cancer Centre (14/178), Prince of Wales Hospital (HREC/14/POWH/261), Sir Charles Gairdner Hospital (2015–048) and the Women's and Children's Hospital (HREC/14/WCHN/113).

Participants and Recruitment

AYA participants were recruited through the five state/territory YCS, where a nominated team member was responsible for identifying eligible AYAs and providing participant information and consent forms. AYAs were eligible to participate in the broader AYA-POST validation study if they were aged between 15 and 25 years, had been diagnosed with any cancer in the preceding 3 months, were receiving treatment (any type) at a YCS-affiliated hospital and were assessed by the recruiting clinician as able to complete the survey (e.g. adequate English proficiency). Eligible young people were invited to the study by a research assistant/nurse at their hospital, who provided them with an invitation letter from the research team and a participant information and consent pack. Consenting young people completed paper versions of the questionnaire pack and indicated whether they were interested to take part in an optional T2 interview a few months after completing the T1 surveys. T2 interviews were conducted by members of the research team over the telephone.

HCPs were eligible to participate if they were employed by the YCS during the data collection period. They were invited to complete the online survey by email.

Measures

AYA Clinical Utility Survey

After completing the AYA-POST at T1, AYAs completed several closed and open-ended questions assessing the tool's clinical utility. These items were adapted from Breen et al.'s (2012) work (see Patterson et al., 2015, for details). This included eight items on the appropriateness, practicability and acceptability of the tool (**Table 1**), which participants responded to using a five-point rating scale (1 = 'strongly agree' and 5 = 'strongly disagree'). AYAs were also asked if the tool covered the main areas they needed (yes/no), and if there were any other questions that should be asked (open-ended).

AYA Service Responsiveness Survey

During the T2 interview, participants responded to seven items about how completing the AYA-POST at T1 may have impacted the care they received from HCPs (**Table 1**) using the same five-point rating scale (1 = 'strongly agree' and 5 = 'strongly disagree'). Three of these items were adapted from Breen et al.'s (2012) work; four additional items were developed to assess whether the AYA-POST facilitated the provision of useful information and referrals, and increased comfort in discussing and seeking help for emotional and psychological needs. Participants were also asked how many referrals they had received for their needs (open-ended).

TABLE 1 | Examples of items assessing appropriateness, practicability, acceptability and service responsiveness of the AYA-POST.

Construct	Example Items
Appropriateness	The tool covered issues that were relevant to me (AYA T1 survey) The tool covered issues I thought were important for AYA cancer patients (HCP survey)
Practicability	The language in the tool was easy to understand (AYA T1 survey) Administering the tool has slowed down or interfered with clinical operations (HCP survey)
Acceptability	I would be happy to complete the tool again as part of my future care (AYA T1 survey) I would be happy to administer the tool to future patients (HCP survey)
Service responsiveness	After completing the tool last time, my medical care team made me aware that help was available if I needed it (AYA T2 survey) The tool helped patients receive appropriate follow-up (HCP survey)

HCP Clinical Utility Survey

HCPs completed an online survey assessing the utility of the AYA-POST. Two open-ended questions asked participants if there were topics not covered in the AYA-POST that were relevant to AYAs, and if there were topics covered in too much detail. HCPs then indicated the extent to which 10 factors (e.g. 'lack of time') were barriers to screening for distress, using a five-point Likert scale from 1 ('not at all a barrier') to 5 ('very much a barrier'). Finally, HCPs who had previously used the AYA-POST rated their agreement with eleven items assessing the appropriateness, practicability and acceptability of the tool (**Table 1**) using a five-point Likert scale from 1 ('strongly agree') to 5 ('strongly disagree'). These items were adapted from previous work on the acceptability of and barriers to distress screening among HCPs (Tavernier et al., 2013; Ristevski et al., 2015; see Patterson et al., 2015, for details).

Data Cleaning and Analysis

Participants who did not complete any clinical utility measures were excluded from analyses. Given the low prevalence of missing data, these responses were not imputed. Responses using the five point *strongly agree—strongly disagree* scale were collapsed into three categories ('strongly agree/agree', 'unsure' and 'disagree/strongly disagree') for ease of interpretation. Responses using the five point *not at all a barrier—very much a barrier* scale were similarly collapsed as: 1–2 = 'not a barrier', 3 = 'somewhat a barrier' and 4–5 = 'barrier'.

Descriptive statistics (frequencies, means and standard deviations) were computed to assess clinical utility (appropriateness, practicability and acceptability) and service responsiveness. Chi-square analyses were used to explore whether perceived clinical utility differed according to AYA age (15–20 years vs. 21–25 years), AYA sex (female vs. male), HCP discipline (nursing vs. allied health; medical professionals excluded) or HCP length of time using the AYA-POST (<3 years vs. >3 years). A Bonferroni correction was applied to correct for the elevated probability of Type I errors when conducting multiple comparisons. The corrected cut-off for significance was $p = 0.006$ for the eight AYA analyses, and $p = 0.002$ for the 21 HCP analyses.

As open-ended responses to questions about items which could be added/removed were few and brief, formal qualitative analysis was not considered appropriate. Instead, commonalities were identified and grouped in order to summarise participant suggestions for item inclusion/deletion.

RESULTS

Participant Characteristics

In total, 118 AYAs (15–25 years, $M = 20.7$ years, SD = 3.2 years; 57 females, 61 male) completed the T1 survey either alone (43.2%) or with family/a partner (27.1%), a HCP (31.4%) and/or another patient (0.8%). Thirty ($M = 22.1$ years, SD = 2.3 years; 17 females, 13 male) chose to complete the T2 interview approximately 3 months later ($M = 86.9$ days, SD = 50.4; range 48–274). Twenty-nine HCPs (medical, nursing and allied health) completed the HCP survey. **Table 2** provides further demographic information about these participants, as well as analyses comparing T2 respondents and non-respondents (T2 respondents were slightly older than non-respondents but did not significantly differ in any other respect).

AYA Perspectives on Clinical Utility

Immediately after completing the AYA-POST at T1, AYAs generally agreed that the tool was acceptable: clear (98%) and easy to understand (97%), relevant (90%) and helpful in communicating emotional needs to their healthcare team (66%). They also reported not needing help to complete the tool (76%). Chi-square analyses indicated there were no evidence of significant differences in ratings by AYA age or sex. **Figure 1** shows the response to all AYA acceptability questions.

Almost all (95%) of AYAs agreed that the AYA-POST had covered all the main areas of their needs. Eight participants reported that they had experienced concerns not included in the NA related to treatment (e.g. delays), social activities (e.g. missing specific hobbies), physical effects (e.g. tinnitus), emotions (e.g. homesickness) or information needs (e.g. next steps for follow-up).

Similarly, some participants' suggestions of additional items which could be included were already included in the NA (e.g. educational and employment concerns) or overlapped with existing items (e.g. sport may be covered by 'missing doing the "normal stuff" with friends'). Participants also suggested heath and healthcare concerns which were not entirely captured by the 'other medical worry' category—for example, relationships with the medical team, concerns about slow healthcare systems.

AYA Reports of Service Responsiveness

At T2, participating AYAs generally agreed that the care they had received had improved since completing the AYA-POST (**Figure 2**): for example, they reported being given useful information (90%) and referrals (90%). Almost all (90%) of these participants had used the services they were referred to, and these reportedly helped them to adjust to their cancer experience (93%). They also reported being more comfortable discussing (90%) and seeking help (73%) for their emotional and psychological needs since completing the AYA-POST. When asked how many referrals they had received since T1, participants reported 2.83 referrals on average (range 0–10, SD = 2.60), with 87% of respondents having received at least one referral.

HCP Perspectives on Clinical Utility

Of the 29 HCPs, 23 reported having used the AYA-POST in their clinical practice; almost half (48%) had been using the tool for over 3 years. HCP ratings of the clinical utility of the AYA-POST (for the 23 who reported previous use) are displayed in **Figure 3**. While HCPs agreed overall that the AYA-POST was acceptable, relevant and feasible, approximately 40% felt it helped them manage patient distress or improve patient care. Chi-square analyses indicated that the distribution of responses did not differ significantly by HCP discipline (nursing

TABLE 2 | Demographics of participating AYAs and HCPs.

Demographic	***n* (%)**		**Test of difference****	
Adolescents and young adults (T1)	**T1 participants (*N* = 118)**	**T2 participants (*N* = 30)**	χ^2	***p***
Sex			1.126	0.289
Female	57 (48)	17 (57)		
Male	61 (52)	13 (43)		
Age			4.893	0.027
15–20 years	55 (49)	7 (24)		
21–25 years	57 (51)	22 (76)		
Cultural and linguistic background*				
Aboriginal and/or Torres Strait Islander	5 (4)	2 (7)	0.585	0.444
Born overseas	15 (13)	2 (7)	1.367	0.242
Speaks another language at home	16 (14)	3 (10)	4.149	0.126
Location			0.209	0.901
Metropolitan	84 (71)	21 (70)		
Rural	28 (24)	7 (24)		
Remote	6 (5)	2 (7)		
Employment (at diagnosis)*			–	–
Working	68 (58)	19 (63)		
Studying	58 (49)	12 (40)		
Home duties	5 (4)	2 (7)		
Volunteering	2 (2)	1 (3)		
On leave	1 (1)	1 (3)		
Unemployed, looking for work	11 (9)	2 (7)		
Cancer types*			–	–
Lymphoma	40 (34)	13 (43)		
Leukaemia	23 (19)	4 (13)		
Sarcoma	21 (18)	3 (10)		
Testicular	15 (13)	5 (17)		
Brain/central nervous system	8 (7)	2 (7)		
Ovarian	3 (3)	0		
Breast	2 (2)	1 (3)		
Colorectal	2 (2)	1 (3)		
Other	13 (11)	2 (7)		
Treatment status			2.014	0.365
Not yet started	12 (10)	5 (17)		
On treatment	103 (87)	24 (80)		
Not sure	3 (3)	1 (3)		
	M (SD), range	*M* (SD), range	*F*	*p*
Age at survey completion (years)	20.7 (3.2), 15–25	22.1 (2.3), 16–25	7.594	0.007
Age at diagnosis (years)	20.1 (3.2), 14–25	21.5 (2.3), 16–25	8.996	0.003
Healthcare professionals	Participants (*n* = 29)			
Discipline				
Medical	3 (10)			
Nursing	15 (52)			
Psychology	4 (14)			
Social work	5 (17)			
Youth work	2 (7)			
Received training on AYA psychosocial distress screening				
At YCS workshop	14 (47)			
On the job	10 (36)			
	M (SD), range			
Time in role (months)	41.7 (41.8), 2–128			

**AYAs could report multiple of these options, if applicable.*
***Between T2 respondents and non-respondents.*

vs. psychosocial) or time using the AYA-POST (<3 years vs. >3 years). Of note, reflecting the perceived feasibility of the tool, HCPs mostly did not perceive that administering the tool slowed down or interfered with clinical processes.

Six HCPs suggested additional items which could be added to the AYA-POST; these included difficulties obtaining financial support from the government, menstrual disruption and social media/technology concerns. No HCPs identified

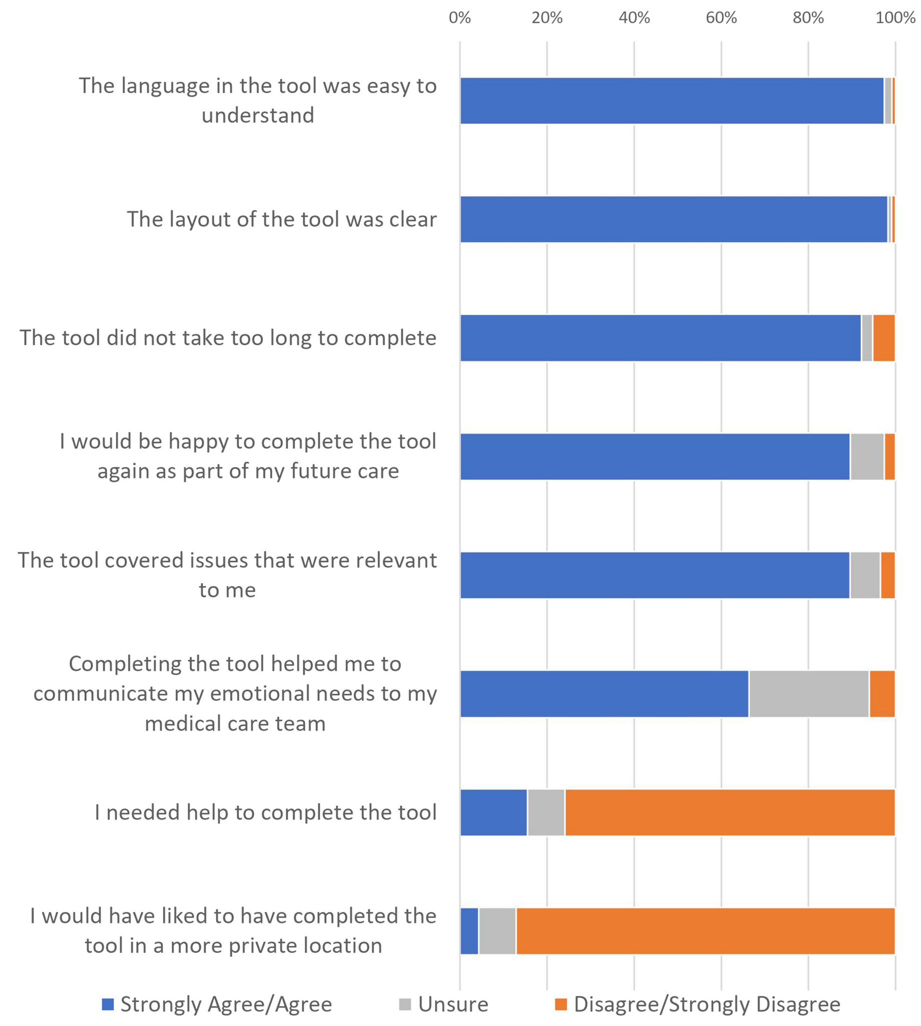

FIGURE 1 | AYA ratings of the acceptability of the AYA-POST.

items that were irrelevant or could be removed from the NA.

HCP Reported Barriers to Screening for Distress

Overall, results indicated HCPs reported few perceived barriers to screening, with the most common being patients were too unwell or distressed (44% barrier and 15% somewhat), and patients being unwilling or reluctant to discuss distress (19% barrier and 41% somewhat). **Figure 4** shows the proportion of HCPs who indicated that each item was a barrier. Chi-square analyses found no evidence that the distribution of responses differed significantly by HCP discipline (nursing vs. psychosocial) or time using the AYA-POST (<3 years vs. >3 years).

DISCUSSION

The findings of this study demonstrate the clinical utility of the AYA-POST, with both AYAs and HCPs rating the tool as broadly appropriate, practicable and acceptable. Additionally, the tool reportedly facilitated communication about emotional and psychosocial needs, and its use prompted referrals which were overwhelmingly experienced as helpful, indicating good service responsiveness. Results also indicated HCPs reported few perceived barriers to screening, with the most common being patients were too unwell or distressed. Together with findings from the international validation study (Patterson et al., 2021b), this study provides strong support for the AYA-POST as a suitable measure for use as standard clinical practice in the biopsychosocial screening of AYAs with cancer, helping to identify distress and unmet needs among patients and facilitating the triaging and tailoring of care. This is a particularly important development, given the absence of validated, population-specific psychosocial measures for this group (Clinton-McHarg et al., 2010; Palmer et al., 2014).

While HCPs agreed overall that the AYA-POST was acceptable, relevant and feasible, around 40% felt the tool helped them manage patient distress or improve patient care and about the same percentage were unsure. Interestingly, these two items

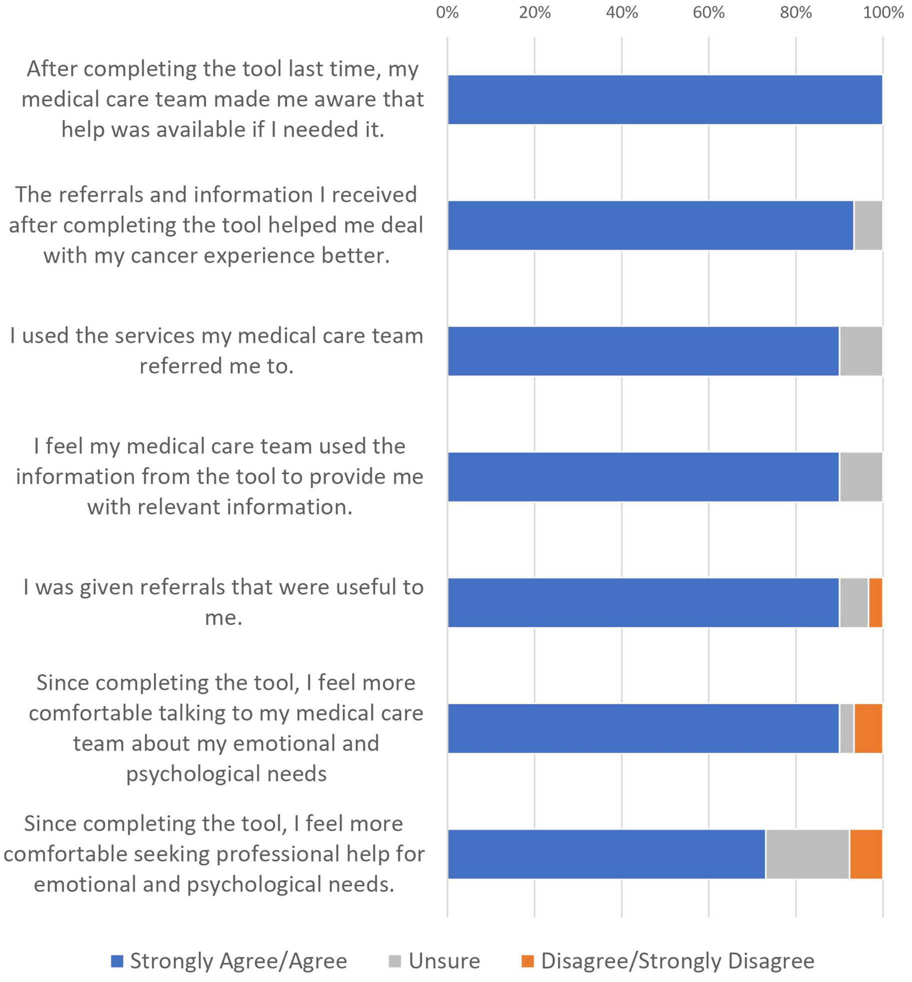

FIGURE 2 | AYA ratings of service responsiveness since completing the AYA-POST.

had the highest 'unsure' ratings by HCPs when assessing elements of clinical utility. In the absence of further information, it is difficult to know why these two items presented the greatest uncertainty for HCPs; importantly, when looking at AYA patients' reported experiences of care these concerns were not evidenced, indeed there was substantial reporting on the benefit of the tool in improving their care and helping them manage their emotions. Further research may be useful to better understand the HCP's responses.

Smart's model of clinical utility defines appropriateness in terms of the efficacy and perceived relevance of a tool (Smart, 2006). While the concurrent validation study confirmed the ability of the AYA-POST to identify patients experiencing clinically significant levels of distress with acceptable sensitivity and specificity using a cut-off score of 5 (Patterson et al., 2021b), this study indicated that both AYAs and HCPs perceived the measure to be effective in terms of facilitating communication about psychosocial distress and needs. Both groups largely agreed that the AYA-POST helped patients to communicate about their needs with HCPs. AYAs additionally noted that it made them more comfortable to talk about their emotional and psychological needs and seek professional help, while HCPs reported that the tool helped with communication and rapport building. While the efficacy of distress screening measures and processes is typically assessed in terms of their performance at identifying and ameliorating psychosocial issues (e.g. Carlson et al., 2012), this indicates a secondary benefit of screening using the AYA-POST in introducing and legitimising discussion of psychosocial issues. This has also been suggested in previous studies of the DT with adults (Dabrowski et al., 2007; Thewes et al., 2009; Johnson et al., 2010) and has the potential to improve engagement with subsequent psychosocial assessment and treatment.

This study also provided evidence of the relevance of the tool to AYAs diagnosed with cancer. For example, almost all AYAs and HCPs agreed that the AYA-POST covered issues thought to be important for this population, and likewise nearly all AYAs reported that the tool covered their main areas of need. Few participants nominated unique concerns which they were experiencing or thought should be included which were

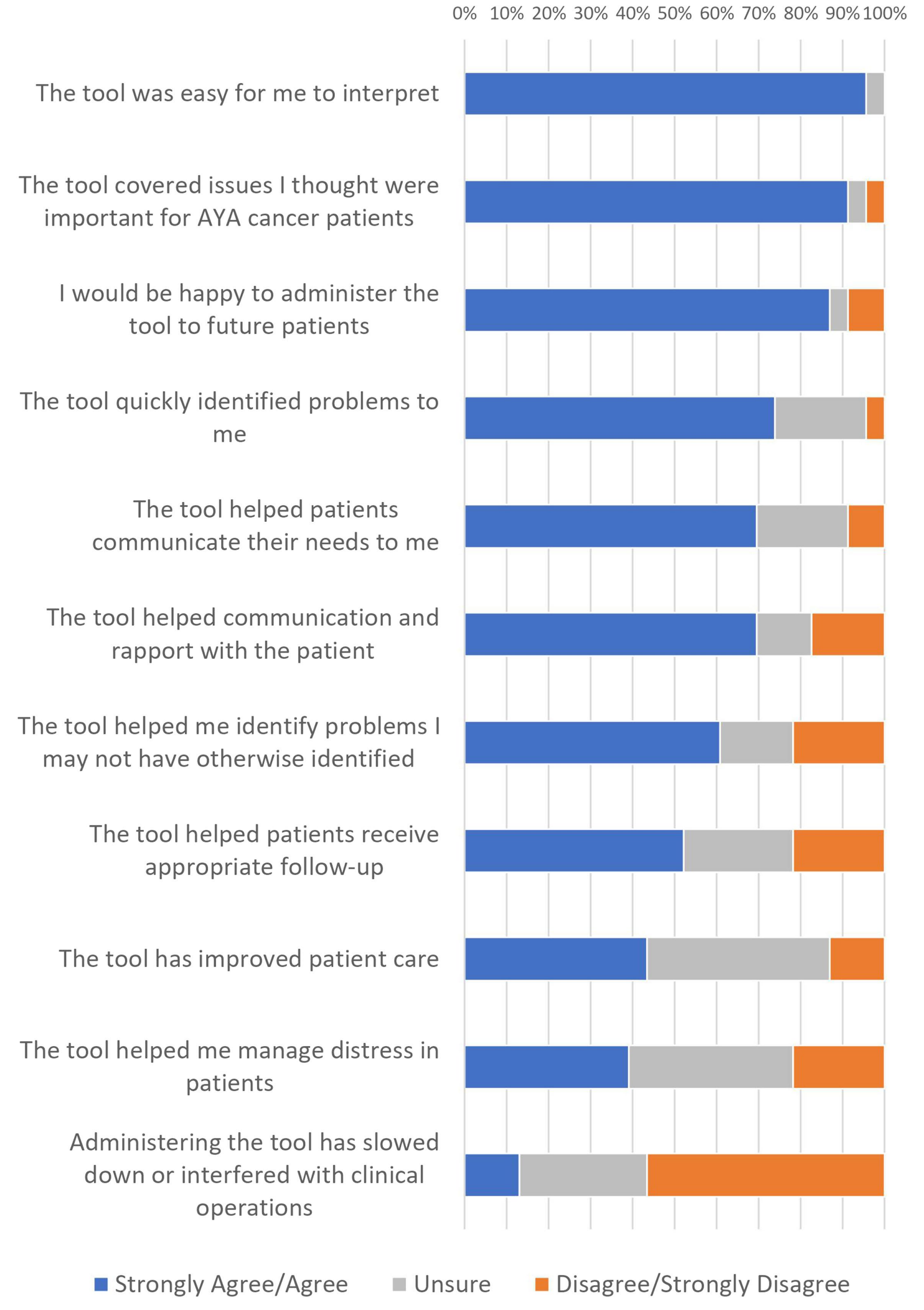

FIGURE 3 | HCP ratings of the clinical utility of the AYA-POST.

not covered to some extent by the existing items, and no AYA or HCP nominated items for removal. That few recommendations for improvement were made indicates that the NA successfully captures the full range of AYA-specific concerns, which provides support for the content validity of the tool (Haynes et al., 1995). This is consistent with findings from the international validation of the AYA-POST, which reported that five of the 10 most commonly nominated issues were AYA-specific additions not present in the adult PCL (Patterson et al., 2021b). Both AYAs with cancer and HCPs specialising in AYA oncology were involved in the development of the AYA-POST (Palmer et al., 2014), and the positive findings around the tool's appropriateness (acceptability and practicability) are a testament to the success of this participatory design approach in ensuring stakeholders' perspectives inform research and service delivery. This is key in ensuring that a psychosocial screening measure is effective.

The practicability of a measure captures the functionality and suitability of materials, as well as whether users have

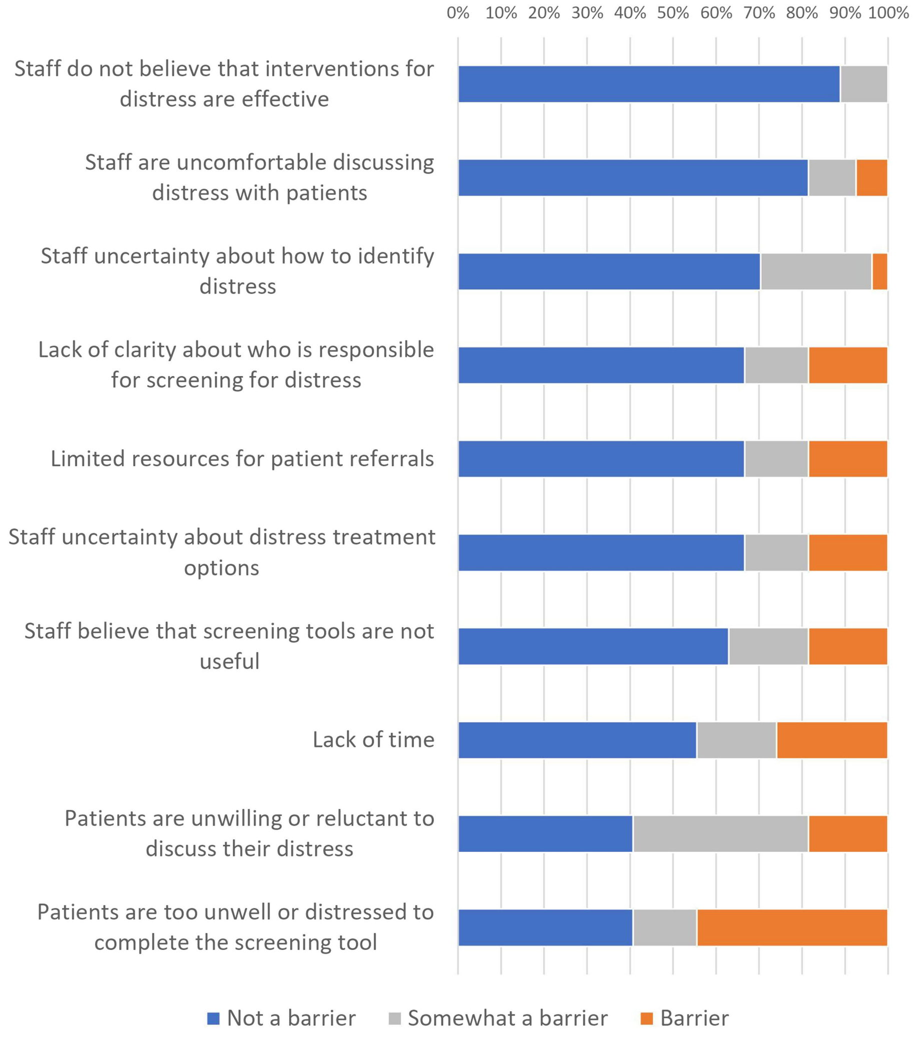

FIGURE 4 | HCP ratings of barriers to use of the AYA-POST.

sufficient knowledge and training to use it (Smart, 2006). The surveyed AYAs almost universally agreed that the language and layout used in the AYA-POST were clear, while only a small proportion reported needing help to complete the tool. HCPs were similarly positive about the practicability of the AYA-POST: the majority of those surveyed agreed that it was easy to interpret and quickly identified problems to them, including problems which they may not otherwise have identified. Likewise, AYAs broadly agreed that the measure did not take too long to complete and few HCPs felt its administration slowed down clinical operations; this echoes previous research which has found that the introduction of standardised psychosocial assessment does not increase consultation times (Engelen et al., 2012) and that clinicians are largely satisfied with the time needed to complete these measures (Teela et al., 2020). These considerations are crucial in selecting an instrument to screen for distress, ensuring that both patients and HCPs can easily understand the measure, minimising the need for further explanation or training. Interestingly, HCP ratings of practicability did not differ between nursing and psychosocial staff; this may be due to the strong holistic focus of the YCS, established protocols around psychosocial care and high uptake of training on distress screening among participants (83%). The AYA Oncology Psychosocial Care Manual offers more detailed recommendations as to how the AYA-POST may be interpreted and implemented as part of a more comprehensive assessment and care pathway (Canteen, 2011), and this may be useful in supporting HCPs with less psychosocial training to use the tool in their work.

The acceptability of the AYA-POST to patients and HCPs is largely reflected in their accounts of the measure's clarity, relevance and helpfulness discussed above; additionally, 90% of AYAs and 87% of HCPs reported that they would be happy to complete or administer the AYA-POST again. These ratings did not differ significantly between female and male AYAs, or between younger and older AYAs; further work may be useful in exploring whether the measure is similarly acceptable to groups underrepresented in this study (and research more broadly) who may have different needs and experiences of care, such as AYAs from culturally/linguistically diverse backgrounds, who are LGBTQI+, or who have disabilities (Wakefield et al., 2013). Of note, this study did not explicitly explore the accessibility of the AYA-POST.

As Carlson (2013) notes, the success of a distress screening process depends not only on the properties of the tool itself, but also on how the health service responds to the results of screening. Previous evaluations of distress screening programmes have shown that screening does not always translate into referral and uptake of psychosocial support services (Carlson, 2013; Mitchell, 2013; Funk et al., 2016), and this lack of follow-up may underlie the limited benefits evidenced for some screening programmes (Carlson, 2013; Mitchell, 2013). By comparison, results from the follow-up AYA survey indicated that the AYA-POST helped to facilitate the provision of appropriate information and referrals to meet patients' needs. The majority of AYAs reported that since completing the tool, their HCPs had made them aware of help available to them and provided relevant information and useful referrals. Uptake of these referrals was reportedly high, more so than previously reported for AYAs [78% (Ellis et al., 2009)] and older adults [30–40% (Ellis et al., 2009; Johnson et al., 2017)]. AYA participants further indicated that these services helped them to better deal with their cancer experience. These results are encouraging and importantly emphasise the associations of targeted referrals and efficient and effective early intervention with administering the AYA-POST. Highlighting these outcomes in training on the tool and institutionalising its use will increase uptake and maximise the benefits it provides for AYAs. It is also worthwhile noting that screening using the AYA-POST may have greater psychosocial benefit for AYA patients being treated within a model of care such as the YCS due to the multidisciplinary nature of YCS teams, together with their strong ties to youth-based community organisations, providing a rich network of internal and external appropriate supports to whom AYAs can be referred to and in a timely manner (Osborn et al., 2019; Patterson et al., 2021a).

Interestingly, the surveyed HCPs were more reserved in their assessment of whether the AYA-POST impacted care: just over half reported that the tool had helped patients receive appropriate follow-up (52%), while 39 and 43% indicated that its use had helped them manage distress and improve patient care, respectively. This discrepancy suggests that HCPs may underestimate the benefits of using a screener like the AYA-POST; being informed of the current results from AYA patients on the usefulness of the tool and subsequent referrals could help HCPs to better understand this.

Finally, overall results indicated HCPs reported few perceived barriers to screening, with the most common being patients were too unwell or distressed (44% barrier and 15% somewhat), and patients being unwilling or reluctant to discuss distress (19% barrier and 41% somewhat). HCP concerns about AYAs' illness, distress and reluctance were not evidenced in the responses of AYAs surveyed here. However, this may be influenced by sampling biases; AYAs' decision to participate in a study on distress screening may be an indicator of their openness to discussing psychosocial issues. Certainly, AYA reluctance to discuss distress has been previously identified as a barrier to accessing psychosocial support, which has been attributed to personal preferences for internalised coping (Holland et al., 2020). It may be that a concise, needs-based measure like the AYA-POST offers a more palatable route to discussing distress for patients who may otherwise be reluctant to engage in interview style assessments. Further research is needed to determine this. System-level barriers around resourcing and responsibility are more frequently reported in the literature (Fradgley et al., 2019; Knies et al., 2019) and suggest a need for services to increase investment in psychosocial staffing to ensure that all AYAs have access to quality care, particularly where patient numbers are expected to increase.

While this study was conducted in the context of the Australian YCS, which is notable for its strong emphasis on age-appropriate, holistic and multidisciplinary cancer care (Osborn et al., 2013; Patterson et al., 2021a), the positive benefits and practice implications discussed throughout have the potential to be similarly realised within international operating environments. It is possible that the positive HCP ratings of the clinical utility of the AYA-POST may to some extent reflect the context of the YCS which places significant emphasis on the psychosocial needs of young people with cancer and has established protocols around the provision of supportive care, and these views may not generalise to HCPs working in non-AYA-specific services or those placing less emphasis on psychosocial care. By contrast, we would expect AYAs' positive views on the AYA-POST to be more broadly generalisable across settings, although we encourage further research exploring its relevance and appropriateness with young people from underrepresented groups who may have different needs and/or experiences of care. We acknowledge however that AYAs who were more ill or distressed, or who were less open to discussing psychosocial concerns, may be underrepresented among participants (particularly among the small number who chose to complete the T2 survey). Indeed, anecdotal accounts from YCS HCPs involved in recruiting AYAs for this study indicated that some HCPs elected not to promote this project to patients they considered too unwell or distressed, meaning that the clinical utility of the AYA-POST among this subgroup of AYAs is less certain. However, as clinician burden made the collection of data on response rates and non-respondents unfeasible, it was not possible to confirm this.

CONCLUSION

Overall, this work indicates high AYA and HCP satisfaction with the AYA-POST, demonstrating its acceptability, practicability and appropriateness in ensuring AYAs with cancer receive appropriate psychosocial care. Combined with concurrent work validating the tool with an international cohort of young people (Patterson et al., 2021b), the study provides strong evidence to support the use of the AYA-POST in psychosocial screening and care provision for these patients, as well as demonstrating the feasibility of using the measure to provide tailored care and referrals. Use of the tool assists in standardising universal screening and referral processes, improving consistency of care (Kim et al., 2018). It can also be useful in detecting psychosocial concerns among the broader AYA cancer population who may not consider themselves to be at risk or who are hesitant to express concerns and support needs themselves, and in detecting issues in domains which may be overlooked, avoided or mistakenly assumed to not be a concern/relevant in non-standardised assessment processes (Skaczkowski et al., 2018).

The AYA-POST can also serve as a useful strategic service planning tool. Identifying commonly reported areas of concern, the data gleaned from its administration can be used to better understand the psychosocial experience of young people with cancer, ensure (or advocate for) sufficient hospital-based services and/or the establishment of clear referral pathways to community-based support to address these issues. Policy makers and health ministries alike can also utilise AYA-POST information in their consideration of developing evidence-based patient-focused models of care for young people with cancer.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material,** further inquiries can be directed to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the ethical approval from the Human Research Ethics Committees at seven lead sites across the country: ACT Health (ETH.11.14.331), Children's Health Queensland Hospital and Health Service (HREC/14/QRCH/374), Northern Territory Department of Health and Menzies School of Health Research (HREC-2014-2,275), Peter MacCallum Cancer Centre (14/178), Prince of Wales Hospital (HREC/14/POWH/261), Sir Charles Gairdner Hospital (2015–048) and the Women's and Children's Hospital (HREC/14/WCHN/113). Written informed consent from the participants' legal guardian/next of kin was not required to participate in this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

PP and FM: conceptualization and project administration. PP, FM, KA, and HB: formal analysis. PP, FM, HB, MO, KM, and AA: investigation. PP, FM, US-D, and AA: methodology. PP and KA: writing—original draft. PP, FM, KA, HB, MO, KM, US-D, KT, MP, and AA: writing—review and editing. All authors contributed to the article and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2022.872830/full#supplementary-material

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Characterizing Early Changes in Quality of Life in Young Women With Breast Cancer

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Introduction: Younger age at diagnosis is a risk factor for poor health-related quality of life (HRQOL) in long-term breast cancer survivors. However, few studies have specifically addressed HRQOL in young adults with breast cancer (i.e., diagnosed prior to age 40), nor have early changes in HRQOL been fully characterized.

Methods: Eligible female patients with breast cancer were identified through our local cancer center. To establish HRQOL, patients completed the Functional Assessment of Cancer Therapy-Breast (FACT-B) around diagnosis and 12 months later. Sociodemographic factors, genetic susceptibility to cancer, tumor- and treatment-related factors, and comorbidities (e.g., depression/anxiety) were abstracted from medical records and the local oncology registry. Mixed-effects models were used to identify changes in FACT-B scores during the first year of treatment and to determine whether any demographic/treatment-related factors modulated changes in scores.

Results: Health-related quality of life in young patients with breast cancer was within normal limits at baseline, with a FACT-B overall well-being score of 108.5 (95% confidence limits [CI] = 103.7, 113.3). Participants reported slight improvements over a 12-month period: FACT-B overall well-being scores increased 6.6 points (95% CI = 2.1, 11.1, $p < 0.01$), functional well-being improved 3.0 points (95% CI = 2.0, 4.1, $p < 0.01$), emotional well-being improved 1.9 points (95% CI = 0.9, 2.8, $p < 0.01$), and physical well-being improved 1.5 points (95% CI = 0.2, 2.8, $p = 0.03$), on average. Participants with anxiety/depression at baseline reported greater improvements in FACT-B overall well-being (change: 12.9, 95% CI = 6.4, 9.5) and functional well-being (change: 5.2, 95% CI = 3.5, 6.9) than participants who did not have anxiety/depression at baseline (change in FACT-B overall well-being: 4.9, 95% CI = 0.2, 9.7; change in functional well-being: 2.3, 95% CI = 1.1, 3.4). Marital status, reconstructive surgery, and baseline clinical staging were also significantly associated with changes in aspects of HRQOL, although their impact on change was relatively minimal.

Conclusion: Young women with breast cancer do not report HRQOL concerns during the first year of treatment. Improvements in HRQOL during the first year of treatment may be attributable to a sense of relief that the cancer is being treated, which, in the short run, may outweigh the negative late effects of treatment.

Keywords: health related quality of life, survivorship, young adult, breast cancer, risk factors

INTRODUCTION

Approximately 5% of new cancer diagnoses in the United States occur in adolescents and young adults (AYA) who are between 15 and 39 years old (SEER, 2022) at the time of diagnosis. Breast cancer accounts for up to 30% of cancer diagnoses among young women (Miller et al., 2020; Scott et al., 2020; Cathcart-Rake et al., 2021) and often presents aggressively with a higher frequency of adverse histopathological characteristics, worse prognosis, and higher risk of recurrence (Gewefel and Salhia, 2014; Johnson et al., 2018). In 2020, the 5-year relative survival rate for young women breast cancer was estimated to be 86% (Miller et al., 2020), making survivorship an important consideration. Research in AYA cancer survivorship has intensified recently (Smith et al., 2013; Nichols et al., 2021), although a considerable amount of what is published about AYA cancer survivorship has been extrapolated from childhood cancer survivor cohorts (Prasad et al., 2015; Jacola et al., 2016; Miller et al., 2020). As a result, survivorship research specific to young women with breast cancer is lagging.

Health-related quality of life (HRQOL), a broad concept encompassing the perceived physical and mental health of individuals (CDC, 2021), is an important endpoint in clinical trials (Haslam et al., 2020). Women with breast cancer reported the highest prevalence of unmet needs in survivorship (Miroševič et al., 2019), underscoring the urgency of continued research on HRQOL in this population. Younger age at breast cancer diagnosis has been identified as a risk factor for reduced HRQOL in breast cancer survivors (Ganz et al., 2003). For instance, Champion et al. (2014) showed that women diagnosed prior to the age of 45 years were more likely to report depressive symptoms, fatigue, and more attention problems than women diagnosed after age 45. Another study reported that female breast cancer survivors who were diagnosed between 35 and 50 years old had a steeper decline in HRQOL in the first 3 years after treatment and recovered more slowly compared with survivors who were diagnosed at an older age (Roine et al., 2021). Notably, while younger age at diagnosis has been established as a risk factor for reduced HRQOL in breast cancer survivors, HRQOL in young adult breast cancer was not specifically addressed. Patients with breast cancer who are in their 20 and 30 s may experience different social and physical challenges than older patients (Quinn et al., 2015), highlighting the importance of studying HRQOL in young women with breast cancer.

A variety of factors besides younger age have been implicated in poor long-term HRQOL in breast cancer survivors. These factors range from somatic predictors (e.g., fatigue, upper extremity lymphedema, obesity, and menopausal symptoms such as hot flashes and sleep disturbances) (Rossi and Pagani, 2017; Saha et al., 2017; Schmidt et al., 2019; Dominici et al., 2021; Jørgensen et al., 2021; Lei et al., 2021; Park et al., 2021) to social factors (e.g., lower socioeconomic status, lack of private insurance, and race/ethnicity) (Ashing-Giwa and Lim, 2009; Derouen et al., 2013; Samuel et al., 2016; Claridy et al., 2018; Dominici et al., 2021) and mental health concerns (Van Esch et al., 2012; Schoormans et al., 2015; Carreira et al., 2018). While HRQOL in breast cancer survivors of all ages may be impacted by these factors, some predictors are more pertinent to younger survivors. For instance, concerns about premature menopause and infertility are more common among survivors diagnosed prior to age 51 and adversely affect HRQOL (Howard-Anderson et al., 2012). Relatedly, young patients with breast cancer typically require intensive therapy due to aggressive tumors that are often diagnosed at a later, more advanced stage (Murphy et al., 2019; Cathcart-Rake et al., 2021). The associated side effects of intense treatment regimens pose a greater risk of poor physical and emotional well-being (Samuel et al., 2016). Dominici et al. (2021) showed that long-term HRQOL was dependent on local therapy strategy in breast cancer survivors who were diagnosed prior to age 40. Young women who underwent more extensive surgeries (e.g., unilateral/bilateral mastectomies) reported lower HRQOL than peers who received breast-conserving therapy.

Finally, most research efforts to date have focused on long-term outcomes (Champion et al., 2014; Cohee et al., 2017; Dominici et al., 2021; Roine et al., 2021). As a result, little is known about HRQOL in the early stages of survivorship. Research conducted in the first year of treatment across a variety of cancers in AYAs identified an unmet need for mental health or fertility counseling services (Quinn et al., 2015), disruptions in work (Schmidt et al., 2019), educational pursuits (Parsons et al., 2012), and parenting (Bailey et al., 2010). The needs of patients and survivors change as they adjust to their cancer diagnosis and survivorship (Costanzo et al., 2007; Husson et al., 2017). Characterizing patterns of HRQOL and factors associated with reduced HRQOL in AYA breast cancer could improve preventative care and patient empowerment (Howard-Anderson et al., 2012; Gewefel and Salhia, 2014).

In summary, there is a knowledge gap regarding early changes in HRQOL in young women with breast cancer, limiting effective surveillance of HRQOL in this population. To address this gap, we aimed to characterize HRQOL in the first year following diagnosis in women with breast cancer who were diagnosed between 18 and 39 years old. We also sought to explore the impact of patient- and treatment-related factors on modulating HRQOL. Given findings of poor HRQOL in long-term breast cancer survivors (Champion et al., 2014; Roine et al., 2021), we hypothesized that HRQOL would decline during the first year of treatment.

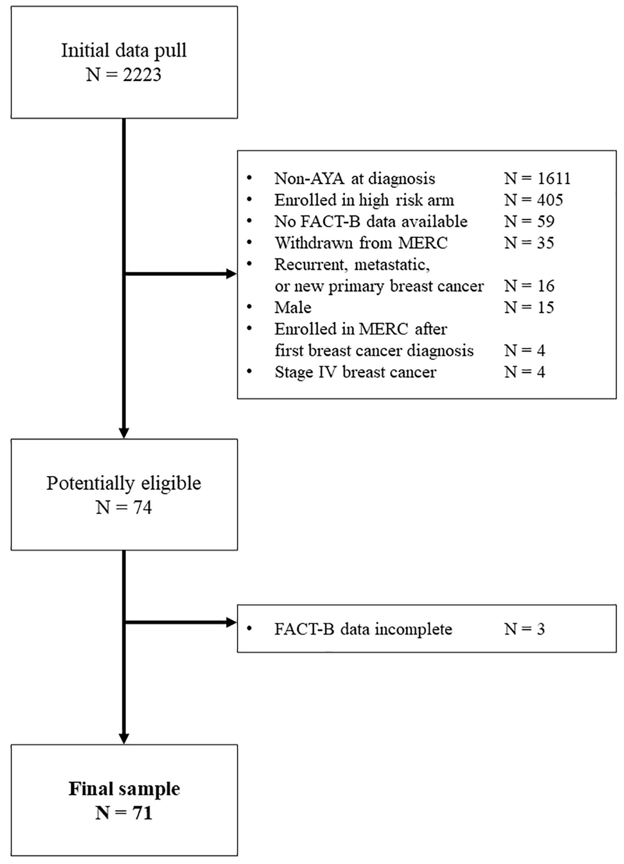

FIGURE 1 | Consort diagram.

MATERIALS AND METHODS

Participants

Participants were identified through the Breast Molecular Epidemiological Resource Core (BMER) data repository at the Holden Comprehensive Cancer Center, University of Iowa Hospitals and Clinics. Patients were eligible to enroll in BMER once they received a definitive breast cancer diagnosis (e.g., primary cancer, recurrence, or metastatic disease) and up until 1 year post-diagnosis. For the current study, patients enrolled in BMER who were female and were diagnosed between ages 18 and 39 were eligible. Those who experienced metastatic and relapsed cancers were excluded, however, of 74 potentially eligible participants, 71 (96%) were included in the final sample (**Figure 1**).

Standard Protocol Approvals, Registrations, and Patient Consents

Participants gave written informed consent prior to enrolling in the BMER study. Directly identifiable information was removed from the data. This project was approved by the Institutional Review Board for Human Subjects Research at the University of Iowa (ID 202106560).

Outcome Measure

Health-related quality of life was ascertained with the Functional Assessment of Cancer Therapy—Breast (FACT-B). This measure is composed of the Functional Assessment of Cancer Therapy—General (FACT-G) (Cella et al., 1993) and 10 additional questions that are specific to a breast cancer subscale score (BCS). The FACT-B is widely used in breast cancer-specific samples and shows high reliability (Brady et al., 1997).

The Functional Assessment of Cancer Therapy—General provides an overall well-being score, as well as scores for physical well-being (PWB), social well-being (SWB), emotional well-being (EWB), and functional well-being (FWB). The FACT-G overall well-being scores range from 0 to 108, with 108 indicating excellent well-being. Since the FACT-B includes the breast cancer-specific questions of the BCS, the maximum value for FACT-B overall well-being is 148. Questionnaires were mailed or emailed to patients prior to their clinic appointment. Responses were recorded on a five-point Likert scale, and higher scores represent better quality of life.

Predictors

Patient Characteristics

Patient variables of interest included (1) sociodemographic factors (i.e., age at diagnosis; insurance status at diagnosis [private/public]; race/ethnicity; and marital status [married/not married]); (2) family history of cancers [no/yes]; (3) tumor- and treatment-related factors (i.e., estrogen receptor status [positive/negative]; progesterone receptor status [positive/negative]; HER2 status [positive/negative]; and fertility counseling [no/yes]); and (4) comorbidities (i.e., body mass index [BMI] and depression/anxiety at the time of cancer diagnosis [no/yes]). Note that insurance status was used as a proxy for socioeconomic status. Further, anxiety/depression was determined by retrieving ICD codes for the presence of anxiety/depression or by identifying whether the individual had an active prescription of antidepressants or anxiolytics at the time of diagnosis.

Medical Parameters

Relevant medical variables were abstracted from medical records and the oncology registry at the University of Iowa Hospitals and Clinics and included the following: (1) clinical staging [0–I/II–III]; (2) laterality [left/right]; (3) surgery type [lumpectomy/mastectomy]; (4) reconstruction surgery [no/yes]; (5) chemotherapy [no/yes]; (6) targeted therapy [no/yes]; (7) radiation [no/yes]; (8) hormone therapy [no/yes]; and (9) ovarian suppression [no/yes].

Statistical Approach

For descriptive purposes, mean baseline scores of participants were plotted with published normative means obtained from the FACT-G in US adults (*n* = 1,075) and adults with cancer (*n* = 2,236; Brucker et al., 2005).

Changes in FACT-B subscale scores were estimated using linear mixed-effects models. Random effects were included to account for the longitudinally correlated nature of repeated

HRQOL assessments at unequal time spacing between visits with a spatial power correlation structure.

Linear mixed-effects models were also used to measure the association between the rate of change in FACT-B subscale scores and baseline clinical, comorbidity, and treatment characteristics. Random effects were similarly included to account for repeated measures with unequal spacing between visits. All statistical testing was two-sided and assessed for significance at the 5% level using SAS v9.4 (SAS Institute, Cary, NC, United States). Plots were generated in R.

RESULTS

Sample

A total of 71 women had completed a FACT-B questionnaire at enrollment (mean 1.5 months from diagnosis) and/or a questionnaire 12 months after enrollment (mean 11.4 months) in which at least one subscale was complete. The median age at diagnosis was 35.0 years (range 22.0–39.0). **Table 1** presents the demographic and treatment information.

Health-Related Quality of Life

Descriptive statistics for FACT-B subscale scores at baseline and at 12 months post-diagnosis are shown in **Table 2**. HRQOL at baseline was within normal limits relative to a normative sample of adults and adults with cancer in the United States (Brucker et al., 2005; **Figure 2A–E**), although young patients with breast cancer reported lower emotional well-being than the reference samples (**Figure 2D**).

Across scales, there was minimal change in scores from baseline to the 12-month assessment. Generally, a slight increase in scores was evident (**Table 2** and **Figures 3A–F**), which reached statistical significance for FACT-B overall well-being (change score: 6.6, 95% CI = 2.1, 11.1, $p < 0.01$; **Figure 3A**), functional well-being (change score: 3.0, 95% CI = 2.0, 4.1, $p < 0.01$; **Figure 3B**), emotional well-being (change score: 1.9, 95% CI = 0.9, 2.8, $p < 0.01$; **Figure 3C**), and physical well-being (change score: 1.5, 95% CI = 0.2, 2.8, $p = 0.03$; **Figure 3E**).

Predictors of Change in Health-Related Quality of Life

Estimated means and rates of change for each predictor across scales are shown in **Supplementary Tables 1–6**.

Anxiety/depression at diagnosis was significantly associated with the rate of change in FACT-B overall well-being, where the anxious/depressed group showed a greater increase in FACT-B overall well-being (change: 12.9, 95% CI = 6.4, 19.5) than the group that was not anxious/depressed (change: 4.9, 95% CI = 0.2, 9.7; **Figure 4A**). A similar trend for anxiety/depression status was observed for functional well-being, with the anxious/depressed group exhibiting a greater increase (change: 5.2, 95% CI = 3.5, 6.9) than the non-anxious/depressed group (change: 2.3, 95% CI = 1.1, 3.4; **Figure 4B**).

Changes in emotional well-being varied significantly as a function of family history of cancer, where those with a family history of cancer exhibited greater improvements in emotional

TABLE 1 | Sample characteristics.

Variable	Levels	Sample $N = 71$
Patient characteristics		
Socio-demographic factors		
Age at diagnosis		**Median (range)**
		35.0 (22.0–39.0)
		Frequency (%)
Race	American Indian or Alaska Native	1 (1.4)
	Asian	3 (4.2)
	Other	3 (4.2)
	White	64 (90.1)
Ethnicity	Hispanic	2 (2.8)
	Non-Hispanic	69 (97.2)
Insurance status	Medicaid	4 (5.7)
	None	4 (5.7)
	Private	62 (88.6)
	Missing	1
Marital status	Married	49 (74.2)
	Not Married	17 (25.8)
	Missing	5
Genetic susceptibility		
Family history of cancer	No	31 (43.7)
	Yes	40 (56.3)
Hormone receptor status		
Estrogen receptor status	Negative	33 (46.5)
	Positive	38 (53.5)
Progesterone receptor status	Negative	36 (51.4)
	Positive	34 (48.6)
	Missing	1
HER2 status	Negative	44 (68.8)
	Positive	20 (31.3)
	Missing	7
Fertility counseling	No	51 (71.8)
	Yes	20 (28.2)
Comorbidities		
Anxiety/depression at diagnosis	No	50 (71.4)
	Yes	20 (28.6)
	Missing	1
BMI		Median (range)
		25.1 (18.4–41.8)
Medical parameters		
Baseline clinical stage	0–II	22 (33.8)
	II–III	43 (66.2)
	Missing	6
Laterality of tumor	Left	39 (54.9)
	Right	32 (45.1)
Surgery type	Lumpectomy	17 (23.9)
	Mastectomy	54 (76.1)
Reconstruction surgery	No	29 (40.8)
	Yes	42 (59.2)
Chemotherapy	No	13 (18.3)
	Yes	58 (81.7)
Targeted therapy	No	51 (71.8)
	Yes	20 (28.2)
Radiation	No	33 (46.5)
	Yes	38 (53.5)
Hormone therapy	No	42 (59.2)
	Yes	29 (40.8)
Ovarian suppression	No	60 (84.5)
	Yes	11 (15.5)

TABLE 2 | FACT-B scores at baseline and at 12 months post-diagnosis.

Covariate	Baseline	Change (12 months–Enrollment)	*P*-value
	Mean (95% CI)	Mean (95% CI)	
Overall well-being	108.5 (103.7, 113.3)	6.6 (2.1, 11.1)	<0.01
Physical well-being	22.6 (21.4, 23.8)	1.5 (0.2, 2.8)	0.03
Social well-being	23.5 (22.4, 24.6)	−0.8 (−1.9, 0.3)	0.13
Emotional well-being	16.0 (14.9, 17.1)	1.9 (0.9, 2.8)	<0.01
Functional well-being	19.6 (18.3, 20.9)	3.0 (2.0, 4.1)	<0.01
Breast Cancer Subscale	27.4 (25.7, 29.1)	−0.1 (−2.1, 1.8)	0.88

well-being (change: 3.5, 95% CI = 2.2, 4.7) than participants who did not have a family history of cancer (change: 0.2, 95% CI = −1.1, 1.4; **Figure 4C**).

Change in the breast cancer subscale scores varied significantly as a function of marital status, where patients who were married exhibited a slight decrease (change: −1.5, 95% CI = −4.1, 1.0), while those who were not married at baseline reported some improvement (3.3, 95% CI = 0.0, 6.7, **Figure 4D**).

Clinical staging (0–I vs. II–III) and reconstruction (no vs. yes) were both associated with change in social well-being. First, patients with stage II–III cancer exhibited a slight decline in social well-being scores (change: −1.7, 95% CI = −3.0, −0.3), while those with stage 0–I cancer showed a slight improvement (change: 1.0, 95% CI = −1.1, 3.1; **Figure 4E**). Likewise, patients who did not undergo reconstruction surgery showed a slight decrease in social well-being (change: −2.5, 95% CI = −4.1, −0.9), while those who had undergone reconstructive surgery reported relatively stable social well-being over the assessment period (change: 0.2, 95% CI = −1.1, 1.5; **Figure 4F**). Results were consistent upon inclusion of both clinical staging and reconstruction in a multivariable model.

DISCUSSION

Young women who were diagnosed with breast cancer in young adulthood report excellent HRQOL at diagnosis and even exhibit some improvement over a 12-month period. Our results suggest deficits in HRQOL may not emerge until later in survivorship. There is strong evidence for impaired long-term HRQOL in breast cancer survivors, particularly those who were diagnosed at a younger age (Kroenke et al., 2004; Howard-Anderson et al., 2012; Champion et al., 2014; Carreira et al., 2018; Maurer et al., 2021; Park et al., 2021). The results from the present study are in line with recent work in early breast cancer (Criscitiello et al., 2021), although others have reported declines in HRQOL during treatment in general cohorts of patients with breast cancer (Schoormans et al., 2015; Maurer et al., 2021). Improvements in HRQOL during treatment have also been reported in pediatric cancer populations who undergo

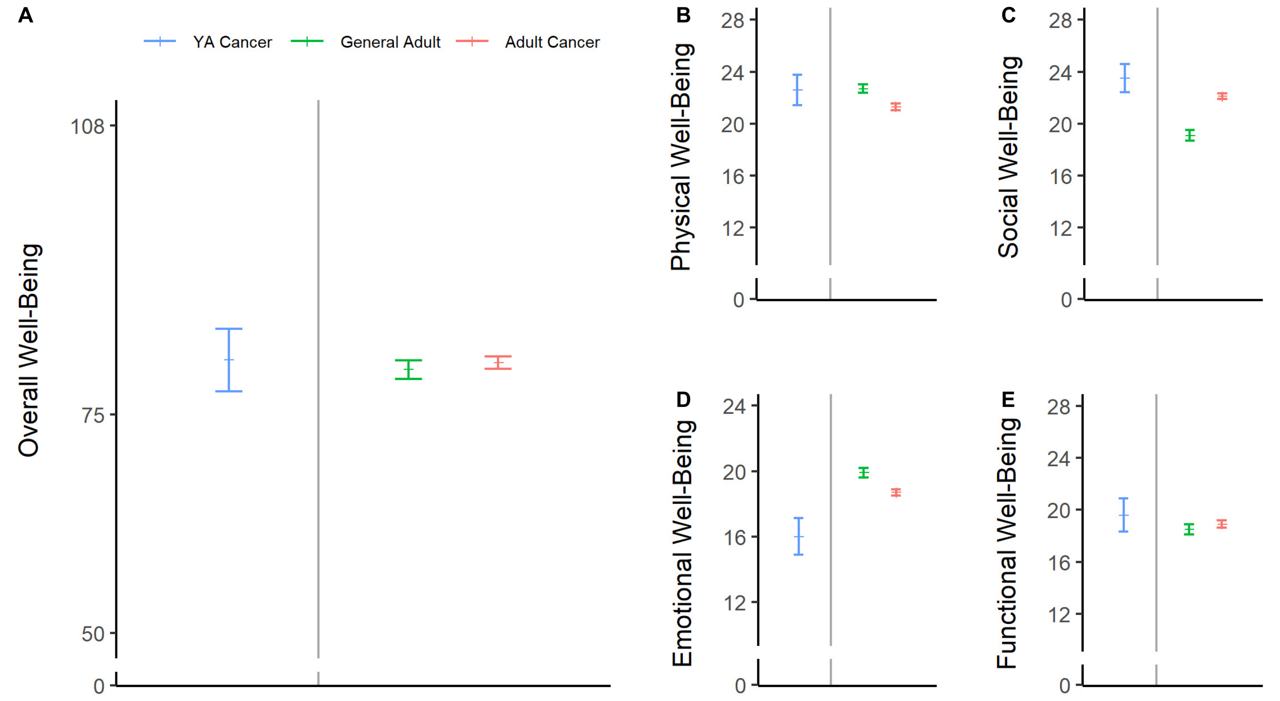

FIGURE 2 | Baseline health-related quality of life (HRQOL) scores in adolescents and young adult (AYA) patients with breast cancer compared with normative data. The sample's mean scores and 95% confidence limits of the means are shown in blue for overall well-being **(A)**, physical well-being **(B)**, social well-being **(C)**, emotional well-being **(D)**, and functional well-being **(E)**. Green lines represent the means of the general US adult population (ages 18–91), and pink lines represent the scores from a sample of adult patients with cancer (ages 18–92) (Brucker et al., 2005).

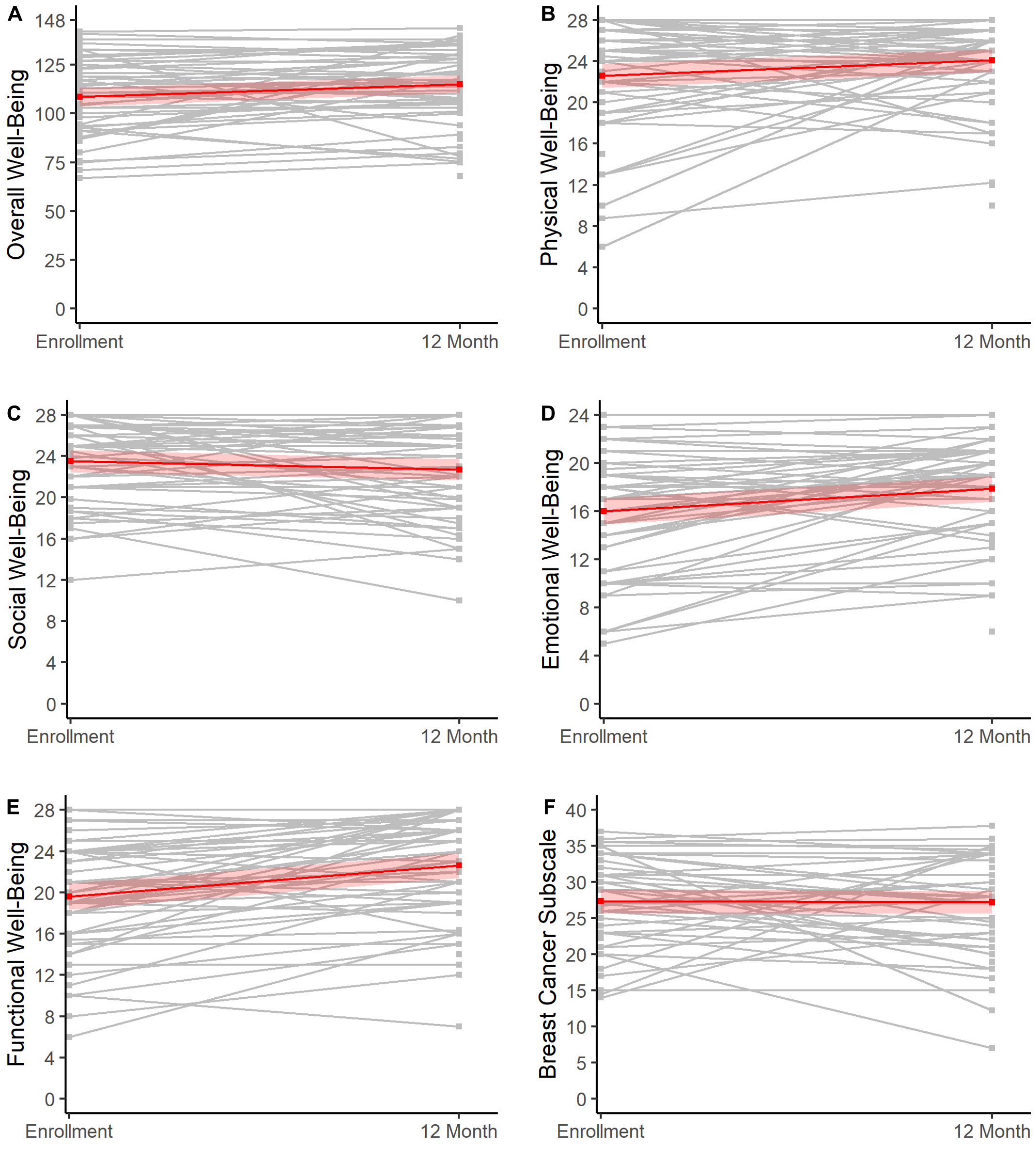

FIGURE 3 | Change in HRQOL from baseline to 12 months post-diagnosis. Changes in HRQOL from enrollment to 12 months post-diagnosis (X-axes) are shown for overall well-being **(A)**, physical well-being **(B)**, social well-being **(C)**, emotional well-being **(D)**, functional well-being **(E)**, and scores on the breast cancer subscale **(F)**. The gray lines represent the individuals, while the red lines show the overall trends across the sample with 95% confidence limits.

intense treatment regimens (Mitchell et al., 2016; Garas et al., 2019). As with long-term breast cancer survivors, pediatric cancer survivors also exhibit poor HRQOL later in survivorship (Kunin-Batson et al., 2014; van der Plas et al., 2020). It seems reasonable to expect that initial improvements in HRQOL are attributable to a sense of relief that the cancer is being treated (Garas et al., 2019), which, in the short run, may outweigh the negative late effects of treatment (Criscitiello et al., 2021). Our results suggest that young adult breast cancer survivors may share this relief to some extent during the first year of treatment and survivorship. However, it is possible that HRQOL diminishes further into survivorship due, in part, to changes in perceived support and increased cumulative burden of treatment-related late effects.

Perceived support from family, friends, and healthcare professionals is an important factor in HRQOL in cancer patients (Usta, 2012; Hurtado-de-Mendoza et al., 2021) and appears to modulate the perceived severity of chemotherapy-related symptoms (Oh et al., 2020). There is evidence to suggest that perceived social support may diminish the further patients that are from diagnosis. A study by Arora et al. (2007) showed that patients with breast cancer reported high levels of emotional support from family and friends at diagnosis; however, emotional support had significantly declined at follow-up. It is possible that

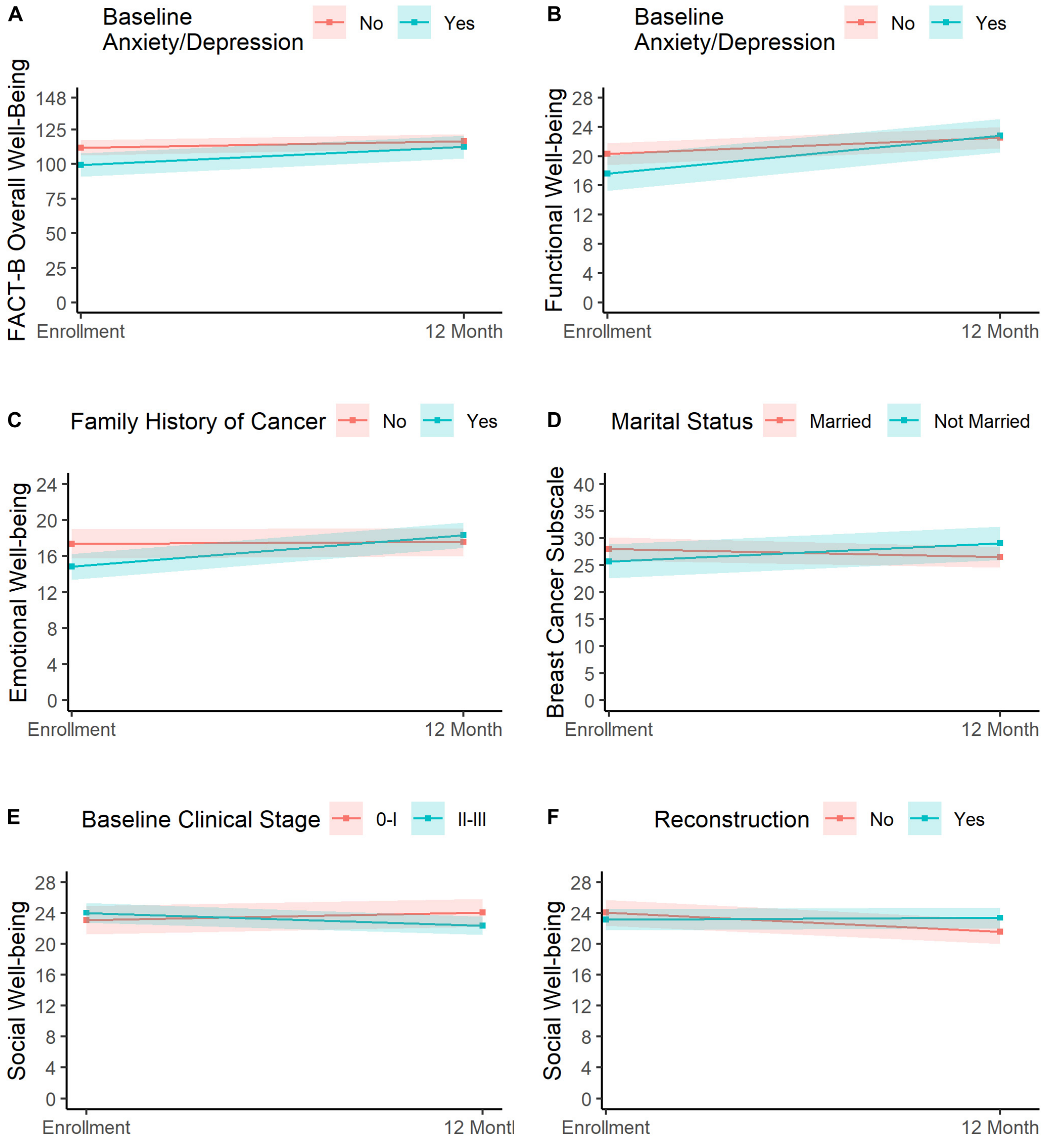

FIGURE 4 | Predictors of change in HRQOL. Change in overall well-being and functional well-being varied significantly as a function of anxiety/depression (**A,B**, respectively), where the anxious/depressed group (blue) showed greater improvement than the non-anxious/depressed group (pink). Change in emotional well-being is shown in panel **(C)**. Participants with a family history of cancer (blue) showed a greater improvement than those without a history of cancer (pink). Panel **(D)** depicts changes in breast cancer scores for married patients (pink) and non-married patients (blue), with the latter showing slight improvements relative to the former. Changes in social well-being varied as a function of baseline clinical stage **(E)** and reconstruction **(F)**. Participants with stage II–III cancer (**E**, blue) exhibited a reduction in social well-being relative to participants with stage 0–I clinical stage (**E**, pink). Finally, participants who did not undergo reconstruction surgery (**F**, pink) reported a slight decline in social well-being relative to participants who did have reconstruction surgery (**F**, blue).

diminishing support contributes to decreased HRQOL further in survivorship of breast cancer survivors. Moreover, young breast cancer survivors (diagnosed < 50 years) with a small social network appear to be particularly vulnerable to low HRQOL in comparison with older survivors (Bloom et al., 2012).

Treatment for breast cancer has been associated with long-term neurocognitive difficulties that are often referred to as "chemo-brain" (Janelsins et al., 2017; Ketterl et al., 2019; Eide and Feng, 2020; Schroyen et al., 2021). Neurocognitive difficulties may encompass memory loss, difficulty concentrating, and other relatively subtle changes that can have a detrimental impact on daily functioning (Chan et al., 2016; Sousa et al., 2020) and are negatively associated with HRQOL (Kunin-Batson et al., 2014; Eide and Feng, 2020; van der Plas et al., 2020; Wagner et al., 2020). AYA cancer survivors across various types of cancers have identified neurocognitive deficits as a key issue (Jim et al., 2018). It is possible that functional difficulties of cancer treatment do not manifest until later in survivorship, even though physiological brain changes are already afoot. In their systematic review, Sousa et al. (2020) reported that there was no evidence for functional change acutely after chemotherapy for breast cancer, even though brain changes were already noted. Others showed that alterations in blood markers of inflammation and neuronal integrity increased the patients with longer breast cancer that are from diagnosis (Schroyen et al., 2021). The notion of "allostatic load" may explain the lag between physiological changes and functional deficits. Allostatic load conveys the cumulative impact of physiological "wear and tear" on the brain and body that eventually results in deterioration in physical and mental health (Suvarna et al., 2020; Schroyen et al., 2021). Thus, a longitudinal assessment of a wide variety of markers indicative of physiological "wear and tear" (e.g., quantitative neuroimaging, blood markers of brain injury, and subtle cognitive decline) is a promising tool to help preserve HRQOL in young breast cancer survivors.

The present study also explored factors that may modulate HRQOL in young women with breast cancer, including sociodemographic factors, genetic susceptibility, hormone receptor status, comorbidities, and treatment-related factors. Anxiety/depression was significantly associated with a change in overall HRQOL and functional well-being. Critically, our target population intersects at two demographics that report an elevated risk of anxiety and depression in cancer survivorship: young age and female sex (Yi and Syrjala, 2017). Approximately 28% of patients had anxiety/depression at diagnosis, which could conceivably have contributed to lower emotional well-being at baseline in this sample (**Figure 2D**). Somewhat surprisingly, patients with anxiety/depression reported overall improvement in HRQOL relative to patients who did not have anxiety/depression at baseline. Young women with anxiety/depression reported lower emotional well-being at baseline and essentially caught up with the non-anxious/non-depressed group 1 year later. One potential explanation is patients with anxiety/depression may have difficulty with emotion regulation in stressful situations (Jazaieri et al., 2015), resulting in greater mood fluctuations than in non-anxious/non-depressed patients. Furthermore, it is possible that the observed improvements in anxiety/depression are related to factors that were not assessed here, such as physical exercise (Patsou et al., 2018). As described above, these improvements may be temporary. Van Esch et al. (2012) reported that anxiety symptoms at breast cancer diagnosis significantly predicted HRQOL 2 years after surgery. Another study showed that the negative impact of depression on HRQOL in breast cancer survivors increased with increased time from diagnosis (Schoormans et al., 2015). Continued surveillance is essential to ensure that these potentially vulnerable, young breast cancer survivors receive the appropriate care (Stafford et al., 2015; Carreira et al., 2018).

Other factors that modulated change in aspects of HRQOL included a family history of cancer, clinical staging at diagnosis, reconstruction, and marital status. Although significant associations were identified, none of these factors appeared to have a clinically meaningful impact on HRQOL during the first year of treatment. For instance, individuals with a family history of cancer reported an average increase of less than four points in emotional well-being. Trends were similar for the other factors that modulated change in HRQOL: Social well-being scores of young women with stage II–III cancer declined with less than two points from baseline to 12 months post-diagnosis, while that of the 0–I group increased one point. Notably, in their research on sociodemographic factors related to HRQOL in breast cancer survivors, Patsou et al. (2018) reported that marital status was not associated with depression/anxiety in survivors age 50 and younger; however, marital status did have a negative impact on depressive symptoms in older breast cancer survivors. These results highlight the possibility that the impact of potential risk factors varies based on age at diagnosis. Collectively, our findings suggest that patient and demographic factors have a minimal impact on changes in HRQOL early in the course of treatment and survivorship in young women with breast cancer.

Limitations

The longitudinal nature of the analyses is a strength of the study, although several limitations warrant mentioning. First, our sample was limited in size, racial diversity, and socioeconomic diversity. These demographic limitations may restrict the generalizability of our findings and underscore the importance of independent replication of the results. Young, African-American breast cancer survivors reported a higher rate of unmet needs, financial distress, and lower physical/functional well-being relative to White breast cancer survivors (Samuel et al., 2016). Yet, African-American patients with breast cancer and survivors are vastly underrepresented in research (Samuel et al., 2016; Nolan et al., 2018), including our own. With a larger and more diverse sample, we can obtain a more holistic and intersectional evaluation of the risk factors that modulate young adult cancer survivorship. Prioritizing research in underrepresented groups will help reduce disparities in HRQOL (Hurtado-de-Mendoza et al., 2021).

Second, breast cancer is exceedingly uncommon among adolescents (SEER, 2022), and our results do not speak directly to adolescent cancer. The AYA age bracket spans a period of dynamic biological and psychosocial changes (Crone and van Duijvenvoorde, 2021). For instance, white matter volume increases rapidly in adolescence, but steady growth of regional white matter is still observed well into the third decade of life (Pomponio et al., 2020). Age-dependent differences in responsibilities, identities, and cognitive abilities also exist in this age bracket, likely resulting in varied survivorship experiences. Increased emphasis on patients' neurodevelopmental stage may contribute to a better understanding of risk factors for reduced HRQOL.

Third, while the FACT-G is established as sensitive to change (Brady et al., 1997), a review by Luckett et al. (2011) notes that research on potential ceiling effects in the FACT-G is lacking. Constructing HRQOL measures with increased sensitivity to change while maintaining brevity for use in clinics can improve clinicians' understanding of patient well-being, screen for patients at risk of decline, and identify promising interventions (Perry et al., 2007).

Relatedly, our results are based on patient-reported outcomes. While these types of assessments remain critical in understanding patient experiences, patient-reported measures have known limitations (McKenna, 2016). One of such limitations is response bias, which can encompass under- or over-reporting of problems (Burchett and Ben-Porath, 2019). As described earlier, broadening the scope and depth of assessments to encompass neuroimaging and blood-based biomarkers may further enhance our ability to appropriately address HRQOL.

CONCLUSION

Health-related quality of life for AYA breast cancer survivors aligns with population norms and remains mostly stable during the first year of treatment. Patient- and treatment-related factors had a limited impact on change in HRQOL during the first year of treatment. While some significant associations were demonstrated (e.g., greater improvement in HRQOL among anxious/depressed patients relative to non-anxious/depressed patients), the clinical significance of these changes remains to be determined. Given that breast cancer at a young age has been clearly associated with poor long-term HRQOL, further study on this population is required to ensure adequate HRQOL is maintained in the long term.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author.

ETHICS STATEMENT

Written, informed consent was obtained from participants before enrollment in the BMER study. Directly identifiable information was removed from the data. This project was approved by the Institutional Review Board for Human Subjects Research at the University of Iowa (ID 202106560).

AUTHOR CONTRIBUTIONS

EP, HA, and SP contributed to the conceptualization, design, and methodology of the study. EP, HA, MC, and SM contributed to the investigation. BL and SM were performed statistical analysis. BL and EP visualized the data. HA and EP wrote the initial draft of the manuscript. SP and EP contributed to funding acquisition. All authors had full access to the data in the study, take responsibility for the integrity of the data and accuracy of the data analysis, revised, read, and approved the submitted version of the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2022.871194/full#supplementary-material

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Psychological Well-Being, Cognitive Functioning, and Quality of Life in 205 Adolescent and Young Adult Childhood Cancer Survivors Compared to Healthy Peers

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The majority of the studies underlined how adolescent and young adult (AYA) Cancer Survivors had no significant differences in their well-being and quality of life compared with a control group of healthy counterparts, although French et al. (2013) found less years of education among cancer survivors. The present study aimed at comparing AYA cancer survivors and a control group of peers who had no history of serious illness, in terms of well-being, cognitive functioning, and perceptions of life. Participants in this study were 205 AYA cancer survivors, 126 males, off therapy from a mean of 10.87 years (SD = 4.91), with a mean age of 18.96 (SD = 3.08), recruited during follow-up visits and healthy counterparts ($n = 205$), matched for age and gender. They all completed self-report questionnaires: Ladder of Life, BSI-18 and Cognitive problems. Paired t test evidenced significant differences between survivors (Mean = 6.19; SD = 2.07) and controls (Mean = 6.88; SD = 2.02) in perceptions of quality of life regarding 5 years before the current time [$t_{(204)} = -3.39$; $p = 0.001$], with a lower level for childhood cancer survivors. Specifically, Hierarchical regression ($R^2 = 0.05$, $p = 0.04$) identified a shorter time since the completion of treatment ($\beta = 0.18$, $p = 0.03$) and a trend of stem cell transplantation experience ($\beta = -0.11$, $p = 0.06$) as factors associated with negative perception of precedent quality of life. The AYA cancer survivors reported lower cognitive difficulties (Mean = 1.46) than controls (Mean = 1.56) [$t_{(204)} = -3.41$; $p = 0.001$]: in memory ($\text{Mean}_{clinical} = 1.32$ vs $\text{Mean}_{control} = 1.50$) [$t_{(204)} = -4.52$; $p = 0.001$], in concentration ($\text{Mean}_{clinical} = 1.36$ vs $\text{Mean}_{control} = 1.54$) [$t_{(204)} = -4.66$; $p = 0.001$] and in mental organization skills ($\text{Mean}_{clinical} = 1.47$ vs $\text{Mean}_{control} = 1.56$) [$t_{(204)} = -2.56$; $p = 0.01$], even if they had a lower educational attainment [$X^2_{(9)} = 131.28$; $p = 0.001$]. They showed similar satisfaction with their psychological well-being and their lives as healthy counterparts, except for past life perceptions associated with the cancer period. Important recommendations for future research and clinical suggestions could be given.

Keywords: AYA cancer survivors, psychological well-being, cognitive functioning, life perceptions, healthy peers

INTRODUCTION

Overview of Psychological and QOL Concerns Experienced by AYA Cancer Survivors

An analysis of the most recent literature presents conflicting results on studies investigating psychological well-being in adolescent and young adult (AYA) cancer survivors.

Some studies, in fact, showed that the quality of life perceived by cancer survivors was good for most of them (Elkin et al., 1997; Zeltzer et al., 2009; Tremolada et al., 2016b). In a group of studies that compared a sample of cancer survivors with a control group of healthy counterparts who had no experience of chronic disease, it was highlighted how survivors perceived themselves more positively than healthy peers. AYA cancer survivors reported fewer depressive symptoms, perceived themselves as more intelligent, and declared having more positive interpersonal relationships (Elkin et al., 1997; Teall et al., 2013). The same researchers suggested that cancer survivors were more likely to portray themselves in a more favorable light than they actually felt. Perhaps the experience of receiving invasive treatments, such as chemotherapy or surgical removal of the tumor mass, might have a positive impact on developing resilience in survivors of childhood cancer. However, also in another recent study (D'Souza et al., 2019), young adult survivors of childhood cancer and their parents did not report increased rates of anxiety or depression compared with their former classroom peers.

In fact, there are many studies that emphasized the positive consequences on psychological well-being of having childhood cancer, especially highlighting the perceived benefits and personal growth that hospitalization experience could offer during adolescence (Kállay, 2006; Tremolada et al., 2016a), also in the hematopoietic stem cell transplantation (HSCT) experience (Tremolada et al., 2018a) and how this construct could also be evaluated by adopting qualitative narrative interviews (Tremolada et al., 2018b).

A second line of studies has a more "pessimistic" view of the psychosocial well-being of these childhood cancer survivors, reporting a preponderance of negative outcomes related to the disease.

Although many surviving patients reported high levels of perceived well-being and satisfaction with their lives, in some situations there were difficulties in accepting and coping with the experience of illness. To confirm these results, the research of Zebrack et al. (2004) and his group of collaborators showed that in the group of survivors, the perception of their own quality of life was significantly lower compared to peers and they reported an increased frequency of symptoms of depression and somatic stress, particularly if they had undergone an intensive chemotherapy. The same author in 2011 found that AYA cancer survivors presented levels of physical and mental health comparable to the average of the healthy population, while their level of perceived discomfort and stress was significantly higher. People who reported feeling more uncomfortable had lower scores on the health scale and reported having more problems with symptoms of anxiety and somatization (but not depression) and a lower quality of life.

Risk Factors of Psychological Concerns in AYA Cancer Patients

In the study of Zebrack and Landier (2011) men, compared to women, generally perceived themselves as healthier although there were no other significant gender differences, not even age at the time of the survey, where no statistically significant differences were reported.

An interesting fact, that emerged from studies on quality of life, was linked to age at the time of diagnosis: those who were older at the time of diagnosis later reported more depressive symptoms and higher levels of stress (Kazak et al., 2010; Zebrack et al., 2012), while younger (< 6 years old), female, and relapsed patients might encounter more life challenges after their disease (Sleurs et al., 2022).

Another study (Kazak et al., 2010) comparing the quality of life and health beliefs of a group of recovered adolescents ($N = 167$) with a control group of peers ($N = 170$) who had never experienced hospitalization, highlighted how survivors perceived themselves as less competent than healthy counterparts in terms of health knowledge: in particular, with respect to their perception of health, with respect to the cognitive skills related to the health topic, and the autonomy they were able to gain from parents. These results could be explained by the negative impact that anticancer treatments and prolonged hospitalization had on cognitive abilities (memory, concentration, and attention) and on ideas about the future of cancer survivors, who experienced it as more uncertain.

Cognitive Impairment in AYA Cancer Survivors and Risk Factors

In a study on the frequency of absences and the school performance of survivors with respect to the perceived quality of life, it emerged that only the perception of one's physical functioning (physical activity, pain, energy) was linked to worse school performance and more frequent absences from school. In fact, these patients declared that they feel more fatigued even when performing daily activities related to domestic tasks or free time (French et al., 2013). Furthermore, this study highlighted how low school performance was predictive of poor university success and difficulties in finding employment. The literature showed that young patients treated with radiation therapy associated with chemotherapy were more at risk for long-term learning difficulties and attention deficit that had a negative feedback in their orientation toward the future. Low school performance was in fact linked to lower self-esteem and worse social skills which in adulthood were reflected in difficulty in finding work, which was higher among survivors than in the general population (Crom et al., 2007). In another recent study (Meernik et al., 2020), financial hardship related to employment disruption among female AYA cancer survivors was found to be substantial.

A recent comprehensive population-based study on long-term survivors of AYA cancer (Dewar et al., 2021) evidenced a higher rate of cognitive dysfunction and psychological distress as compared to the general population. Among cancer survivors, those reporting cognitive dysfunction had greater likelihood of having psychological distress. Similarly, cancer survivors with more severe psychological distress had greater likelihood of reporting cognitive dysfunction.

Research Gaps and Objectives

The literature highlighted how, at present, the results of studies on the quality of life of patients who survived cancer were contradictory, especially if AYA cancer survivors underwent hematopoietic stem cell transplantation (HSCT) (Reinfjell et al., 2017). It was possible to find two opposing views: a number of studies report adverse outcomes (Rourke et al., 2007; Zeltzer et al., 2008; Tai et al., 2012), others conclude that the quality of life and psychosocial adaptations were satisfactory for most of the recovered patients (Elkin et al., 1997). The conflicting results of studies on the quality of life in patients with cancer could be partially explained by the different diagnoses in the study populations, the time elapsed since diagnosis, the methods, and the diversity of tools for evaluating the dependent variables.

There is a lack of studies on the life perceptions and hopes in AYA cancer survivors and also on their cognitive functioning perceptions that could be independent from their real academic performance.

In Italy there are limited studies conducted on this topic and that adopted control groups of healthy peers. To the best of our knowledge, there is only one Italian study (Tremolada et al., 2020) aimed to understand the possible differences in well-being, cognitive, and life perceptions in adolescents in treatment for leukemia with healthy peers. This study adopted the same questionnaires and, surprisingly, found that healthy counterparts have a better perception of current life, but reported a lower hope score, more anxiety symptoms, and more perceived cognitive problems than patients. A possible protective resource could be identified in hopefulness, predictive of a good health-related quality of life (HRQoL) in the near future (Rosenberg et al., 2018). This resource was correlated with a positive sense of wellbeing and commitment to treatment, and also improved coping and self-esteem, especially in females (Cantrell and Lupinacci, 2004). The main purpose of the research is to investigate the patient's level of well-being both on a physical-functional and psychosocial level and to identify and investigate the possible long-term consequences of the disease and the impact this may have in everyday life. Evaluation of the psychological well-being of patients supports outpatient monitoring activity related to the physical functional area, as required by the specific therapeutic protocols for each pathology.

The first group of hypotheses aims to determine if there are significant differences in well-being, life satisfaction, and cognitive functioning in AYA cancer survivors based on sociodemographic and disease parameters. We expected a lower well-being and life satisfaction in females, in older adolescents, those with fewer years off therapy, and experience of stem cell transplantation.

The second group of hypotheses aims to verify whether there are statistically significant differences between the two groups in terms of psychological well-being, life perceptions, cognitive functioning and higher educational level obtained. Based on the existing literature, we expected to find a similar trend in the two groups for psychological well-being and life perceptions. Regarding achieving higher educational qualifications and cognitive functioning perceptions, we hypothesized a worse trend for the AYA cancer survivors.

MATERIALS AND METHODS

Participants

All eligible survivors who attended the Pediatric Hematology-Oncology Clinic of the University of Padua in the period 2015–19 were asked to participate in this project. Eligibility criteria included being treated for cancer before the age of 18, at least 5 years having passed since the last day of therapy, and being currently 15–25 years old. We excluded survivors treated for central nervous system tumors, those with learning or sensory problems or genetic syndromes, and those who were unable to complete the questionnaires independently.

The participants in the control group (n = 205) met the following eligibility criteria: no history of life-threatening or chronic illness or injury and absence of learning or sensory problems and other pathological aspects. The healthy peers in the control group of were enrolled in secondary schools, youth groups, and university faculties in the same region as the patients (Veneto, northeast of Italy).

Procedure

Ethics approval was obtained from the Hospital of Padua committee considering it as an observational and spontaneous study. The day before the follow-up appointment at the Day Hospital of the Clinic, the clinical psychologist telephoned each survivor to explain the study and obtain participation consent for the next day. If the survivor was less than 18 years old, the parent was contacted before the psychologist called the participant. Upon their arrival at the clinic, survivors received a packet that included information about the study, a consent form, and a collection of questionnaires. The consent form was completed by the AYA participants, or in the case of those younger than 18 years of age, by their parents. Participants returned the questionnaires within the following 2 weeks in stamped addressed envelopes or electronically using a protected online site.

The oncologists who followed the patients and conducted the initial query in the database to identify eligible patients abstracted the necessary data from the patients' medical records. Medical data extrapolated included: date and type of diagnosis, therapy protocol involved, age at diagnosis, hematopoietic stem cell transplantation (HSCT yes/no, type and date), date of stopping therapy, years from stop therapy, relapses (yes/no and when) and surgical intervention (yes/no and when).

Measures

Most of the instruments used for this investigation were derived from the Childhood Cancer Survivor Study (CCSS), a cohort of 27 centers in the United States and Canada that assembled survivor samples that were sufficiently large and diverse enough to allow investigators to investigate delayed effects of treatment.

Ladder of Life (CCSS)

Participants had to evaluate, using a 1–10-point scale, the quality of their current life (Current life score), the quality of their life 5 years before their disease (Past life score), and how satisfied their life will be in the future (5 years after the child's diagnosis) (Future life score). There were no cut-offs available that could be used to interpret the scores, but the scale could be easily interpreted using the following rule: an insufficient score was identified in the range 1–5; a sufficient score corresponded to 6–7; and a good score was 8–10. With this instrument, we could obtain information about individual perception of the past, the present, and the future. It had been administered to 118 Italian mothers of patients with cancer, demonstrating good global internal consistency (Cronbach's alpha = 0.73) (Tremolada et al., 2012).

Brief Symptom Inventory 18

The Brief Symptom Inventory 18 (BSI-18) consisted of 18 items grouped into three dimensions of six items, serving as a screening for depression, somatization, and anxiety. Respondents were asked to comment on how they felt in the last 7 days, and each item was rated on a 5-point Likert scale from 0 (not at all) to 4 (extremely) (Derogatis, 2000). BSI-18 was used to assess psychological outcomes in long-term survivors of childhood cancer (Axia et al., 2006; Recklitis et al., 2006) and in mothers of patients under treatment for leukemia (Tremolada et al., 2013). Brief Symptom Inventory 18 ha been administered to 118 parents of patients with cancer, demonstrating good internal consistency for both the global score (Cronbach's alpha = 0.92) and specific domains (Depression: alpha = 0.84; Somatization: alpha = 0.83; Anxiety: alpha = 0.83). Also, for this study, the psychometric parameters were good attesting at alpha values between 0.73 and 0.82.

Scale of Cognitive Problems

This is a 25-item questionnaire, used in another Italian study (Tremolada et al., 2013) that investigated the presence and intensity (range from 1 = "never a problem" to 3 = "often a problem") of cognitive problems shown by AYA cancer survivors dealing with their disease experience in the last 2 weeks. The Cognitive Problems Scale has been administered to 118 Italian parents of patients with cancer, demonstrating good global internal consistency (Alpha = 0.89). A Varimax rotated confirmatory factor analysis, explaining a good proportion of the total variance (56.63%), identified five subscales. Memory (five items; alpha = 0.78); Mental disorder (eight items; alpha = 0.82); Labile mood (three items; alpha = 0.75); Impulsivity (four items, alpha = 0.73); Concentration (five items; alpha = 0.67); these five dimensions could be combined into a total score, Cognitive Problems score (25 items; alpha = 0.89). The Cognitive Problems Scale was derived from the CCSS battery, and the larger purpose of this measure was to assess the frequency of possible cognitive problems that may arise in people who were under huge stress. In this study, the alpha values showed a moderate psychometric reliability between 0.55 and 0.70.

Socio-Economic and Medical Data

Each participant filled out a sociodemographic questionnaire with questions about their highest year of schooling, their mother's and father's education, their perceived economic situation, their type of home situation, their romantic relationship, and their type of employment.

Statistical Methods

A chi-squared test in crosstabs was used to estimate the possible sociodemographic differences between the two samples and to better understand the comparability of the samples. The two groups were matched according to gender and age, and a file was created with the matched clinic and control pairs. Descriptive measures of central tendency and variability were calculated for all relevant variables and comparisons were made between the two groups. Inferential comparisons were made between cancer survivors and control samples using a paired sample *t*-test. We ran preliminary Pearson bivariate correlations and hierarchical regression analyses to find the possible significant associations/predictions between the examined variables. Statistical significance was evaluated at the nominal level of $p = 0.05$, with adjustments for multiple comparisons, after controlling the normal distribution of the test scores.

RESULTS

Characteristics of Study Cohort

Altogether, 230 of the 325 eligible survivors were informed about the study and contacted by telephone, prior to a follow-up visit. The 95 eligible survivors who the researchers did not reach in the study had changed phone number, moved from their original residential location, had no check-ups during the research period, or had no more follow-ups in the clinic. Informed consent and completed questionnaires were received from 222 individuals (response rate = 96.5%). We have no data from the eight patients who refused to participate, so any comparison between the two groups was not possible. Of these 222 survivors, 205 were matched with participants in the control group. All patients were Caucasian with a mean age of 18.96 (SD = 3.08). We matched only 205 patients with the control group because only 205 patient–healthy peer pairs with the same gender and year of birth were possible. **Tables 1**, **2** illustrate the sociodemographic and medical information of the participants and their families.

Demographic and Schooling Level Comparability Between AYA Cancer Survivors and Controls

Comparative analyses of the different areas measured by the questionnaires used in the study were carried out on the final sample made up of 205 AYA out-of-therapy and healthy peer pairs. AYAs were paired considering gender and current age: It was possible to form 79 pairs of women and 126 pairs of men, whose average age was 18.96 years (SD = 3.08).

TABLE 1 | AYA cancer survivors' demographic and disease characteristics.

Characteristic		Survivors (N = 205)		Controls (N = 205)		χ^2; p
		Frequency	**%**	**Frequency**	**%**	
Mean age (Years)	**18.96 (3.08)**					
Gender	Males	126	61.5	126	61.5	ns
	Females	79	38.5	79	38.5	
Education	0–8 years of schooling	90	43.9	101	49.3	$\chi^2_{(16)} = 31.59$; $p = 0.0001$
	9–13 years of schooling	92	44.9	74	36.1	
	> 13 years of schooling	14	6.8	30	14.7	
Relationship status	Engaged	60	29.3	81	39.5	ns
	Single	95	46.3	118	57.6	
	Not reported	50	24.4	6	2.9	
Employment (N = 78)	Looking for a job	19	9.3	8	3.9	ns
	Part-time	14	6.8	23	11.2	
	Full-time	44	21.5	16	7.8	
	Not working	128	62.4	158	77.1	
Diagnosis	Leukemias	79	38.5			
	Lymphomas (Hodgkin and non-Hodgkin)	65	31.7			
	Solid tissue	55	26.8			
	Other	6	2.9			
HSCT	No	177	86.3			
	Yes	28	13.7			
Age at diagnosis, Mean years (SD)	7.09 (4.38)					
Years from end of therapy. Mean (SD)	10.87 (4.91)					

Legend: Solid tissue included the following diagnoses: Hepatoblastoma, Hodgkin lymphoma, Langerhans cell histiocytosis, neuroblastoma, bone tumor, ovarian tumor, rhabdomyosarcoma, retinoblastoma, soft tissue sarcoma, Wilms tumor. HSCT, hematopoietic stem cell transplantation.

There were differences between AYA cancer survivors and controls only in terms of educational level [$X^2_{(9)} = 131.27$; $p = 0.0001$] (**Table 1**). Cancer survivors had significantly less schooling years than controls. Specifically, in our data 14.7% of the participants in the control group have a bachelor's or master's degree, compared to 6.8% of the cancer survivors out of therapy. Furthermore, at the same age, a higher percentage of recovered young people (21.5%) claimed to be full-time workers than their peers in the control group (7.8%).

Regarding the family characteristics of the two groups (**Table 2**), they are quite similar to each other, except for a statistically significant difference in the level of education of the mother [$X^2_{(16)} = 31.584$; $p = 0.01$] and of the father [$X^2_{(16)} = 29.939$; $p = 0.02$]. As can be seen from **Table 2**, the mothers and fathers of the AYA in the control group (68.7 and 54.1%, respectively) have a higher school diploma or degree than the mothers and fathers of the AYA in the out-of-therapy group (40.0 and 47.9%, respectively).

The Associations of Sociodemographic and Disease Parameters With AYA Childhood Cancer Survivors' Well-Being

There were no statistically significant gender differences compared to the other variables in the scores obtained in the BSI-18 and Ladder of Life instruments. A statistically significant difference appeared on the mood subscale [$t_{(202)} = -2.51$; $p = 0.01$; $d = 0.43$; CI 95%: −0.29/−0.12] of the Problem Scale questionnaire in which women (mean = 1.64; SD = 0.51) scored higher than men (mean = 1.47; SD = 0.47). This indicated that women have more mood-related problems, which limited them in managing daily problems.

In the control group gender differences were identified for the BSI-18 depression scale, for past Ladder of Life perceptions and for some Problem difficulties scales (Total, Disorganization, Concentration). **Table 3** shows these results.

There were no significant differences on these scales according to age, while a significant positive association was found between arousal symptoms and schooling years ($r = 0.17$, $p = 0.02$).

A series of Pearson's correlations were run to understand the possible significant associations between the scales of psychological well-being, satisfaction with life and problem difficulties and the parameters of the disease (that is, years off-therapy, age at diagnosis, HSCT yes/no of hematopoietic stem cells transplantation). Quality of life perception scores regarding 5 years before the current time were associated with age at diagnosis ($r = -0.15$; $p = 0.02$), with years off-therapy ($r = 0.22$, $p = 0.001$), and with the presence/absence of HSCT ($r = -0.13$, $p = 0.04$). A hierarchical regression model ($R^2 = 0.05$, $p = 0.04$) identified a significance of less years off-therapy ($\beta = 0.18$, $p = 0.03$) and a trend of significance of presence of HSCT ($\beta = -0.11$, $p = 0.06$) as factors that could negatively influence the quality of life perception regarding 5 years before the current time in AYA cancer survivors.

Psychological Well-Being and Life Satisfaction Comparability Between AYA Cancer Survivors and Controls

The third area concerned the psychological functioning and the perception of one's current life perception, and the first hypothesis, specifically, provided that there are no differences between the two groups considered regarding the perception of one's psychological functioning and life perception. The

TABLE 2 | Demographic characteristics of the AYA cancer survivors' families.

Characteristic		Survivors (*N* = 205)		Controls (*N* = 205)		χ^2; *p*
Mother's education	5 years of schooling	11	5.4	8	3.9	$\chi^2_{(9)} = 131.28$; $p = 0.01$
	8 years of schooling	90	43.9	50	24.4	
	13 years of schooling	66	32.2	89	43.4	
	Degree	16	7.8	52	25.3	
	Not reported	22	10.7	6	2.9	
Father's education	5 years of schooling	17	8.3	11	5.4	$\chi^2_{(16)} = 29.94$; $p = 0.02$
	8 years of schooling	67	32.7	46	22.4	
	13 years of schooling Degree	74	36.1	81	39.5	
	Not reported	24	11.8	38	18.6	
		23	11.2	29	14.1	
Economic situation perceived	Low	19	9.3	25	12.2	ns
	Medium	85	14.5	82	40	
	High	84	41	97	47.03	
	Not reported	17	8.3	1	0.5	
Home situation	Rent home	11	8.3	11	5.4	ns
	Home ownership with mortgage	37	32.7	46	22.4	
	Home ownership without mortgage	124	36.1	81	39.5	
	Other	19	11.8 11.2	38	18.6	
	Not reported	23		29	14.1	

TABLE 3 | Means and SD of depression score, past life perceptions and problem scale scores comparing males and females.

	t(df)	*p*	*d*	Interval confidence 95%	Gender	Mean	SD
BSI depression	−2.15 (203)	0.02	0.31	−0.41/−0.02	Females Males	0.99 0.77	0.78 0.65
Past life perception	1.72 (203)	0.04	0.25	−0.07/1.04	Females Males	6.62 7.11	1.85 2.14
Total Problem difficulties	−2.76 (203)	0.003	0.41	−0.21/0.03	Females Males	1.63 1.50	0.32 0.32
Mental disorganization	−2 (203)	0.02	0.56	−0.2/−0.001	Females Males	0.38 0.36	0.03 0.04
Difficulty concentrating	−2.34 (203)	0.01	0.5	−0.26/−0.02	Females Males	0.44 0.42	0.04 0.04

first analyses were carried out on the scales of the BSI-18 questionnaire and on the questions relating to the Life perception scores, aimed at investigating this type of variable. The analyses did not reveal statistically significant differences related to the psychological well-being experienced, thus confirming the starting hypothesis.

By means of the *t*-test for paired samples, a statistically significant difference emerged in the quality of Life subscale related to the 5 years before the current time [$t_{(202)} = -3.39$; $p = 0.001$] (**Table 4**). The recovered AYA reported to be less satisfied With their precedent quality of life (mean = 6.19; SD = 2.07) than those in the control group (mean = 6.88; SD = 2.02).

The fact that no statistically significant differences emerged in the other two scales of the instrument allowed us to confirm the hypothesis mentioned above.

Perceived Problem Functioning in AYA Cancer Survivors Compared With That in Controls

The last hypothesis, according to which patients off therapy reported greater cognitive difficulties than their peers in the control group, was investigated by taking into account the data collected with the Problem Scale questionnaire. This questionnaire evaluated the perception of one's own general cognitive functioning and the presence of specific difficulties such as mnemonic, attentional, and mental organization.

From the analyses carried out (*t*-test for paired samples), the means that were statistically significant in comparison between the two groups are the following:

- Difficulty in solving general problems [$t_{(204)} = -3.410$; $p = 0.001$], for which the highest scores were detected for the control group (mean = 1.57; SD = 0.33) compared to the group out-of-therapy (mean = 1.46; SD = 0.30);
- Disorganization [$t_{(203)} = -2.557$; $p = 0.01$], in which the control group (mean = 1.56; SD = 0.37) scored higher than the group of Recovered young people (mean = 1.47; SD = 0.34);
- Difficulty concentrating [$t_{(202)} = -4.666$; $p = 0.0001$] for which higher scores are recorded for the control group (mean = 1.54; SD = 0.44) compared to off-therapy group (mean = 1.36; SD = 0.35);

TABLE 4 | Means and SD of life perception and problem scale scores comparing clinic and control group.

	t (df)	*p* value	Confidence intervals 95%	*d*	Group	*N*	Mean	SD
Current life perception	1.24 (203)	0.21	−0.11/0.51	0.15	Clinic	204	7.10	1.83
					Control	204	6.90	1.39
Past life perception	−3.39 (203)	0.001	−1.08/−0.28	0.40	Clinic	203	**6.19**	2.07
					Control	203	**6.87**	2.02
Future Life perception	1.46 (198)	0.14	−0.07/0.49	0.14	Clinic	199	8.29	1.60
					Control	199	8.07	1.38
Total Problem difficulties	−3.41 (204)	0.001	−0.16/−0.044	0.32	Clinic	205	**1.46**	0.29
					Control	205	**1.56**	0.33
Impulsivity	−0.78 (204)	0.43	−0.10/−0.05	0.08	Clinic	205	1.69	0.47
					Control	205	1.73	0.49
Labile Mood	0.013 (203)	0.99	−0.09/0.09	0.14	Clinic	204	1.53	0.49
					Control	204	1.53	0.48
Mental disorganization	−2.56 (203)	0.01	−0.16/−0.02	0.28	Clinic	204	**1.47**	0.34
					Control	204	**1.56**	0.37
Difficulty concentrating	−4.66 (202)	0.0001	−0.25/−0.10	0.45	Clinic	203	**1.36**	0.35
					Control	203	**1.54**	0.44
Memory difficulties	−4.52 (203)	0.0001	−0.26/−0.10	0.47	Clinic	204	**1.32**	0.34
					Control	204	**1.50**	0.47

Bold values indicate the significative mean differences comparing clinic and control group.

- Memory difficulty [$t(203) = -4.522$; $p = 0.0001$] in which the non-therapy group (mean = 1.32; SD = 0.34) scored lower than the control group (mean = 1.50; SD = 0.47).

Table 4 shows these results.

DISCUSSION

Brief Summary of Main Findings

The main purposes of this study were to investigate the psychological well-being, cognitive problems, and life perceptions of AYA childhood cancer survivors, and to identify the illness or sociodemographic factors associated with these outcomes.

We intended to compare the self-reported quality of life and experiences between survivors and matched controls. The main findings of this study are: (1) the lower educational attainment reported by AYA cancer survivors and by their parents comparing with healthy peers, even if recovered patients recognized less cognitive problems; (2) a good current and future quality of life perceptions in AYA cancer survivors compared with healthy peers, even if they reported a worse quality of life in the 5 years before the current time; (3) a good psychological well-being in recovered patients comparable with that of healthy peers; (4) the identification of risk factors for cognitive and psychological difficulties such as female gender, experience of HSCT, less years off therapy.

Cognitive Functioning and Educational Attainment of AYA Cancer Survivors Compared With Healthy Peers

Cancer survivors had significantly lower educational attainment than controls. Our results were in line with the international literature, according to which a large proportion of patients out of therapy tended to terminate their studies earlier than their healthy pairs (Crom et al., 2007; French et al., 2013). The differences between cancer survivors and healthy peers in parents' level of education could be a limit for the pairings, since it was not possible to control it, but it may also be a result of the impact of the patients' illness on the educational and career prospects of the parents. This will be better investigated in the future with the recruitment of other control samples.

However, regarding cognitive functioning, AYA in the out-of-therapy group surprisingly perceived themselves in a more positive light, in both domains, than their peers in the control group. Higher scores corresponded to more cognitive problems. These findings did not support our original hypothesis which presumed that survivors of cancer might perceive more cognitive problems than peers. In fact, the collected data showed that they perceived themselves better than the adolescents and young adults in the control group. On the one hand, compared to healthy pairs, they declared that they have less cognitive difficulties, specifically in terms of concentration and memory, and related to mental disorganization. These data could be explained, in part, by considering the results that emerged from the comparison of the sociodemographic variables of the two groups. The out-of-therapy group had a lower educational qualification than the control group: among survivors, fewer young people had a university degree and more were already in full-time jobs. Perceiving oneself as having less difficulty in cognitive functioning could perhaps be explained by the fact that the AYA cancer survivors, whose studies had been interrupted encountered fewer cognitive problems compared to their peers in the control group, who were more committed in their education and, consequently, more concerned about this area. Another possible confirmation of what has been said so far comes from the results of the analysis of the sociodemographic variables of the families of the two groups. Parental couple of the off-therapy group had lower educational attainment than that of the control group. Probably this variable could also have influenced the choice to stop studies earlier in the young survivors, choosing to follow the parental model.

A further explanation could be due to the fact that the tools used were self-report questionnaires that detect not so much the actual cognitive abilities, but the personal perception that one has of them. It is possible that as a result of the difficult past, cancer survivors out of therapy were led to see themselves in a more positive light, sometimes overestimating their skills.

Quality of Life Perceptions in AYA Cancer Survivors Compared With Healthy Peers

The presence of significant differences between the two groups could be expected: Life perception could be worse in the AYA cancer survivors' group due to the possible long-term consequences of the disease, which is particularly burdensome. However, a good current life perception was shown in the clinic group despite the past traumatic experiences in childhood.

Perhaps, it was possible to advance the hypothesis that these cancer survivors continually compare their current state of health with their difficult past, so much so that they recognized a clear improvement over past years. For this reason, they tended to declare themselves satisfied with their life and cognitive functioning, even overestimating themselves. These results are in line with some studies conducted internationally (Elkin et al., 1997; Teall et al., 2013), according to which survivors were more inclined to represent themselves in a more favorable light than they really felt. Perhaps having been subjected as patients to invasive treatments, such as chemotherapy or surgical removal of the tumor mass, not only taught them to survive but also to overcome and develop in the best possible way, fighting adversity with more determination and valuing all the personal and environmental resources available to them. This phenomenon was related to the concept of post-traumatic growth, an important issue for AYA childhood cancer survivors (Tremolada et al., 2016a).

Confirming the consistency of these results, a significant difference emerged between the two groups in the area of satisfaction with the last five-years of life. Patients out of therapy declared themselves less satisfied when looking back on past experiences than healthy counterparts. These results were likely to be in line with the painful experience of illness experienced by the young people out of therapy in their past, which probably marked them compared to their peers who had never experienced with this traumatic past.

In fact, this particular group of adolescents in the past had faced challenges that left them very debilitated, and this is undoubtedly a differentiating element.

However, despite the memory of the disease still present, these AYA probably aspired to look at their present and future with positivity. Our data seemed to go more in the direction of other research (Elkin et al., 1997; Zeltzer et al., 2009; Teall et al., 2013), according to which cancer survivors, while admitting some difficulties, feel psychologically satisfied and confident for their future development.

Psychological Well-Being in Recovered Patients Comparable With That of Healthy Peers

Regarding mental health status, in some studies a more negative profile emerged in AYA childhood cancer survivors, who declared themselves more tired in their cognitive tasks and as having self-esteem difficulties (Tremolada et al., 2017), having more difficulties at school both from the point of view of performance, with greater absences and the possibility of failing, and from the point of view of achieving higher qualifications (Crom et al., 2007; French et al., 2013; Tremolada et al., 2016b). These data could be explained by considering that the young people could not yet have gained all the resources to cope with the long-term effects that invasive therapeutic procedures had on their well-being and cognitive functioning.

However, the results obtained from the comparison between the two samples of the present research were encouraging and not in line with that part of the literature that described AYA cancer survivors as less satisfied with their lives and as having more limitations than their healthy counterparts.

From the point of view of psychological functioning, no significant differences emerged between the two groups that do not perceive themselves as different with respect to anxiety levels, physical problems, and the presence of possible depressive symptoms. A similar result was also obtained in preadolescents and adolescents during the acute therapy phase (Tremolada et al., 2020).

Both groups were equally satisfied with their current and future life perceptions: these data were comforting since it would seem that the history of illness, undoubtedly painful and difficult to process, had not left negative marks on the lives of these young people who were preparing to face their future with optimism.

Risk Factors for Cognitive and Psychological Difficulties

Some studies highlighted the presence of gender differences among young people out of therapy in terms of perception of their health. In particular, recovered women report higher depressive and emotional symptoms (Zeltzer et al., 2009; Zebrack and Landier, 2011). Our results did partially confirm the presence of gender differences, as the literature tended to indicate. In particular, man and women did not differ in their life perceptions and well-being, but only women reported greater difficulties related to mood. On the other hand, this gender difference was more marked in the control group, where females declared more depressive symptoms, negative life perceptions and several cognitive difficulties such as difficulty concentrating and disorganization. Analyses were also conducted to evaluate differences based on age, but no significant differences emerged in any of the variables in question: even the international literature, which investigated possible differences within the off-therapy group only, did not report any age differences. To sum up possible reflections on this topic, it was possible to advance the hypothesis that the experience of lived illness in some way contributed to make this particular group of participants homogeneous regarding their way of perceiving psychosocial health and well-being both for age and also for gender.

Worse perceptions of life in the precedent 5 years were influenced by fewer years off therapy and the presence of hematopoietic stem cell transplantation. All these disease parameters could be identified as risk factors in determining negative life perception during the treatment period.

Strengths and Limits

This study has some merits, but also some limitations.

We would like to highlight a few strengths of this study. First, we included age- and sex- matched peers as the comparative group. Both survivors and peers were asked if they had any previous hospitalization due to a serious illness before participating in the study. Second, the size and the relative homogeneity of the sample of who participated in the research were really good. Even when controlled for, the medical variables appear to have a limited association with aspects related to the psychosocial well-being of recovered patients and adolescents. Third, both groups received the same complete set of questionnaires that investigated the concept of psychological well-being and life perceptions in a multidimensional way, making it possible to develop complex health profiles.

The study has also some limitations. First, our obtained results were not always homogeneous and coherent with the literature on this topic. Probably, this non-homogeneity could be explained by the fact that, very often, in cancer studies is not usual to find a distinction between different types of cancer in pediatric age, or because uneven control groups were used (healthy pairs not matched by controlling sociodemographic variables or comparing groups such as healthy siblings, groups of patients with other pathologies, etc.) that did not always reflect the socio-demographic characteristics of AYA childhood cancer survivors. Second, the fact that the data were collected exclusively in a region of Northern Italy makes it difficult to generalize the data to the entire national context, due to social and environmental variables that could significantly affect the results collected. Third, the exclusive use of self-report questionnaires allows the rapid collection of a large quantity of information, but which risks being negatively influenced by some variables that are difficult to control, such as social desirability, with the result that collected data do not always reflect reality.

CONCLUSION

Highlighting the most problematic areas could help health professionals to propose psychological and/or psychotherapeutic interventions to those patients and their families found to be most at risk and to those who explicitly request it. For example, a clinical suggestion could be to strengthen the schooling activities during the cancer treatment and in the off-therapy time to improve AYA cancer survivors' educational attainment. It could be useful also to set up cognitive and psychological interventions especially for women, or for those that had a HSCT experience and that were nearer to the stop therapy time. The worse quality of life perceptions related to the experience of the cancer disease could be ameliorated by adopting more recreational, social and educative activities for children and adolescents during the cancer therapies.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Hospital of Padua Committee. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

MT and SB: conceptualization and methodology. MT and LT: formal analysis. MT: investigation, data curation, and writing—original draft preparation. AB: resources. LT and SB: writing—review and editing. MP and AB: visualization. SB and MP: supervision. SB: project administration. All authors have read and agreed to the published version of the manuscript.

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The Perceived Impact of COVID-19 on the Mental Health Status of Adolescent and Young Adult Survivors of Childhood Cancer and the Development of a Knowledge Translation Tool to Support Their Information Needs

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Background: Adolescent and young adult (AYA; 13 to 39 years) survivors of childhood cancer may be especially vulnerable to physical health and mental health concerns during the pandemic. We investigated the impact of COVID-19 on the mental health status of AYA survivors (Aim 1) and shared tailored, evidence-based health-related information on COVID-19 (Aim 2).

Methods: Between May and June 2020, participants completed a cross-sectional online survey assessing their cancer history, current mental health status, and their COVID-19 information needs.

Results: Ninety-four participants (78 females, 13 males, 2 non-binary) with a mean age of 26.9 years (SD = 6.2) were included in the final sample. Participants reported residing from 10 countries and 94% identified as White. Nearly half of the participants (49%) described their mental health status as worse now than before the pandemic. Thirty-nine participants (41%) that indicated their current mental health status was tied to fears/worries about their past cancer and treatment experienced a higher level of anxiety and PTSS than those who did not report the same. Most participants (77%) had not received any information related to the potential risks of COVID-19 and expressed an interest in receiving this information. In response, an infographic detailing recommended strategies for coping with mental health problems in the pandemic, along with preliminary study findings, was developed.

Discussion: AYA survivors reporting their mental health status was linked to their past cancer experienced poorer mental health. There is a value to educating survivors on their potential health risks, but accounting for their perceived mental health vulnerabilities should be considered when disseminating knowledge. The use of an infographic is a unique contribution towards the development of innovative and personalized means of sharing health education to this vulnerable yet resilient group. This research on the mental health status of AYA survivors very early in the pandemic informs continued initiatives investigating the rapidly changing nature of how COVID-19 may impact AYA survivors today and in the future.

Keywords: pediatric oncology, psychosocial oncology, adolescent and young adult cancer, survivors of childhood cancer, mental health, COVID-19

INTRODUCTION

Coronavirus disease 2019 (COVID-19) is an infectious disease caused by a severe acute respiratory syndrome coronavirus 2 (SARS-CoV2; World Health Organization, 2020). As of January 2022, there are over 364 million cases and 5.63 million deaths confirmed worldwide (WHO Coronavirus (COVID-19) Dashboard, 2021). Many individuals infected with COVID-19 experience a range of mild to moderate respiratory symptoms and can recover without medical intervention. Those with underlying medical comorbidities or immunocompromised individuals are among the most vulnerable populations that may be at a greater risk for developing serious illness or dying from COVID-19 (Coronavirus (COVID-19) and the Immunocompromised, 2021).

Due to advances in medical treatment, there is a growing prevalence of survivors of childhood cancer around the world. In fact, 5-year survival of children and youth with cancer is beyond 80% in most European and North American countries (Robison and Hudson, 2014). Further, there is an estimated >500,000 survivors of childhood cancer in North America (Robison and Hudson, 2014). The most common forms of childhood cancer include leukemias, brain cancers, and solid tumour (Steliarova-Foucher et al., 2017; Siegel et al., 2021) Survivors of childhood cancer may be especially susceptible to the impacts of COVID-19 (Forster and Schulte, 2021) due to chronic health conditions, known as late effects, that stem from their cancer treatment (Schulte et al., 2020). These include health concerns that are known to increase risk of a more severe course of COVID-19, including pre-existing cardiac issues, pulmonary disorders, obesity, and diabetes (Neville et al., 2006; Brabant et al., 2012; Ward et al., 2014). Psychological late effects, including anxiety, depression, and post-traumatic stress symptoms (PTSS), are also common in survivors of childhood cancer (Lown et al., 2015; Brinkman et al., 2018; Nathan et al., 2018), and predisposes these individuals to mental health vulnerabilities.

Adolescents and young adults (AYAs) survivors of childhood cancer (henceforth referred to as AYA survivors) have distinct needs. It can be challenging for AYA survivors to meet their health care needs and navigate survivorship (Barr et al., 2016) because this is a critical developmental period during which major changes take place in their living arrangements, health care needs, and transition from pediatric to adult care (Marjerrison and Barr, 2018). Prior to the COVID-19 pandemic, AYA survivors reported experiencing poorer quality of life, including physical and mental health, when compared to the general population (Schulte et al., 2021).

There is emerging research on the negative impact of COVID-19 on the mental health of the general population (Cullen et al., 2020; Pfefferbaum and North, 2020; Usher et al., 2020), as well as those from vulnerable groups (Nearchou et al., 2020). Several studies have explored the psychosocial effects of the COVID-19 on those living with and beyond their cancer, highlighting a greater prevalence of mental health concerns than individuals without a cancer history as well as an enduring sense of fear and worry about their potential health risks (Wang et al., 2020; Islam et al., 2021; Page et al., 2022). Of note, the pandemic has drastically changed the access to and model of delivery of follow-up care for survivors (replacing face-to-face appointments to virtual health), which has implications for the surveillance of their health and psychosocial needs (McLoone et al., 2020). Further, quarantine and social isolation (Pahl et al., 2021) may contribute to feelings of isolation and loneliness (e.g., Brinkman et al., 2018) that further compromise one's mental health (e.g., Nearchou et al., 2020).

AYA survivors may be at a greater risk of mental health concerns given their health history and perceived vulnerabilities. Indeed, a mixed-methods study conducted in the United States at the outset of the pandemic found that AYA survivors reported a high level of anxiety regarding their health and that of their family, feelings of isolation, and worries about employment status (Shay et al., 2021). In another international study comprised of AYA patients and survivors, more than 50% of respondents expressed an interest for information tailored to their needs in coping with the pandemic (Košir et al., 2020). More recently, a study investigating sources of COVID-19 information use by AYA survivors with cancer living in Canada revealed a preference by AYA survivors to seek information through social media and websites of cancer organizations (Yan et al., 2021). Collectively, existing research highlights a need to support AYA survivors in their coping with their mental health and an urgency to deliver health information to this group, likely through innovative and digital methods. Building on these efforts, incorporating the voices and lived experiences of AYA survivors in the generation and dissemination of health information is critical and has been lacking from published studies.

Today, 9.9 million vaccine doses aimed to target COVID-19 have been administered globally [WHO Coronavirus (COVID-19) Dashboard, 2021]. The mental health status of AYA survivors may be different now than early in the pandemic. However, with the emergence of new SARS-CoV2 variants, coupled with changing public health responses around the world, AYA survivors continue to grapple with a great deal of uncertainty related to their potential risks and management of COVID-19. In particular, the mental health of AYA survivors may be compromised in this quickly evolving context. Therefore, a complete assessment of the impact of COVID-19 on the mental health status of AYA survivors, along with the dissemination of tailored, evidence-based information on coping with mental health during COVID-19 in an accessible manner, is necessary to support the on-going well-being of this at-risk and vulnerable group.

CURRENT RESEARCH

The overarching goal of the current research was to identify the impact of COVID-19 on the mental health status of AYA survivors during May and June 2020 and determine their information needs living in the pandemic, particularly given their prior diagnosis and treatment exposure. This goal was carried out through the following aims:

Aim 1a

To explore the perceived impact of COVID-19 on the mental health status of AYA survivors, including ratings of anxiety, depression, and PTSS.

Aim 1b

To determine differences in the mental health status of AYA survivors who reported that their mental health status was associated with fears/worries about their past cancer and treatment, with those who did not report the same.

Aim 2a

To describe the COVID-19-related information needs of AYA survivors.

Aim 2b

To disseminate an infographic outlining our research and evidence-based coping strategies for COVID-19 specific to survivors of childhood cancer.

MATERIALS AND METHODS

Patient and Public Involvement

We reported the background, aims, methods, and results of this study based on the checklist from the Guidance for Reporting Involvement of Patient and the Public Short Form (GRIPP2-SF; Staniszewska et al., 2017). Specifically, the COVID-19 and Childhood Cancer study is a patient-oriented research project and thus developed in collaboration with our program's patient partners to identify priority areas for our research objectives. Patient partners collaborated on the study design, recruitment, data collection, interpretation of results, and knowledge dissemination.

Participants

Participants were recruited as part of a larger study on COVID-19 and childhood cancer. AYA survivors were identified as individuals who were: (1) diagnosed with cancer under 21 years of age; (2) more than 5 years from diagnosis and/or more than 2 years from cancer treatment completion, consistent with the definition established by Children's Oncology Group Long-Term Follow-Up guidelines (Childrens Oncology Group, 2018); (3) currently between the ages of 13 and 39 years of age (National Cancer Institution, 2022). No other restrictions were placed on eligibility to maximize the representativeness of the sample and ecological validity of the findings.

Recruitment

Participants were recruited through a variety of sources, including social media (e.g., Twitter), community organizations (e.g., Kids Cancer Care Foundation, Childhood Cancer Survivor Canada), and convenience sampling through our patient partners. Ethics approval was obtained by the Health Research Ethics Board of Alberta—Cancer Committee (HREBA.CC-20-0151). Data were collected between May and June 2020.

Procedure

This study employed a quantitative, cross-sectional design. Participants completed a survey with questions pertaining to their cancer history, current mental health status, and their understanding of COVID-19-related information. The survey was administered through REDCap™, a secure online platform affiliated with the tertiary care pediatric hospital where the research was based.

Measures

Mental Health

Standardized measures of anxiety, depression, and PTSS were administered as an index of survivors' current mental health status. Each measure is described in detail below.

Anxiety

Anxiety was measured using the anxiety subscale from the Patient Reported Outcomes Measurement Information System (PROMIS Profile-29; e.g., Cella et al., 2007). The PROMIS

Profile-29 assesses anxiety, depression, fatigue, sleep disturbances, peer relationships and cognitive function, pain interference and pain intensity. This measure has been validated in pediatric oncology for 8 to 17 years (e.g., Hinds et al., 2013). Participants were asked to rate 4 items assessing symptoms of anxiety over the past week (e.g., "I felt worried") on a five-point Likert scale from 1 "*never*" to 5 "*always*." Scores range from 4 to 20 with higher scores reflecting greater severity of anxiety. Standardized scores were computed by summing the responses scores on all items to generate a total raw score, which was then converted to a *t*-score with a mean (*M*) of 50 and standard deviation (SD) of 10 based on a US general population. T-scores can be interpreted as follows: < 55: *none* to *slight*; 55–59: *mild*; 60–69: *moderate*; 70+: *severe* levels of anxiety (American Psychiatrists Association, 2022). Internal consistency for this sample was good ($\alpha = 0.89$).

Depression

As with anxiety, depression was measured by a subscale from the PROMIS Profile-29 (e.g., Cella et al., 2007) as described above. Participants were asked to rate 4 items assessing depression symptoms (e.g., "I felt sad") in the past week on a five-point Likert scale from 1 "*never*" to 5 "*always*." Scores range from 4 to 20 with higher scores reflecting greater severity of depression. Standardized scores were computed by summing responses the scores on all items to generate a total raw score, which was then converted to a *t*-score based on the same norm referencing as the anxiety subscale of the PROMIS Profile-29. Score classification (*none to slight, mild, moderate, and severe*) was based on the same criteria as the anxiety subscale. Internal consistency for this sample was excellent ($\alpha = 0.93$).

Post-traumatic Stress Symptoms

PTSS were assessed using two measures. The Child Post-Traumatic Stress Disorder Symptom Scale for DSM-5 (CPSS-5—Self Report Version for DSM-5; ISTSS, 2022) was administered to adolescent survivors. The CPSS-5 is a 27-item self-report measure that assesses PTSD symptoms experienced by children ages 8 to 17 years over the past month. Participants are asked to rate the frequency of PTSS experienced using a five-point Likert scale from 0 "*not at all*" to 4 "*6 or more times a week/almost always*" (e.g., "I have bad dreams or nightmares"). The total severity score ranges from 0 to 80 and was computed by summing the ratings of the first 20 items. Higher score reflecting higher severity of PTSS, and a cutoff score of 31 can be used to identify a probable PTSD diagnosis in children (Foa et al., 2018). Separately, seven items assessing impairment of endorsed symptoms on daily functioning are summed to indicate an impairment score that ranges from 9 to 28. Internal consistency of this sample was excellent ($\alpha = 0.97$).

The PTSD Checklist for DSM-5 (PCL-5; Weathers et al., 2013) was administered to young adult survivors. The PCL-5 is a 20-item self-report measure that assesses PTSD symptoms experienced over the past month by adults 18 years and older. Participants are asked to rate the extent to which they are likely to experience PTSS using a five-point Likert scale from 0 "*not at all*" to 4 "*extremely*" (e.g., "I am bothered by repeated, disturbing, and unwanted memories of the stressful experience"). Total score was computed by summing the scores of all items. Scores range from 0 to 80 with higher score reflecting higher severity of PTSS. A cutoff score between 31 and 33 is indicative of a probable diagnosis of PTSD. In this study, we referred to a lower cutoff point to increase detection of possible cases of PTSD. Internal consistency of this sample was excellent ($\alpha = 0.95$).

Perceived COVID-19 Impact

Participants responded to 6 questions regarding the perceived impact of COVID-19 on their current mental health status and daily living circumstances. These items were developed by the research team with experts (researchers, health care providers) in pediatric psychosocial oncology and in collaboration with our patient partners to shape research priorities. An example item regarding the perceived impact of COVID-19 on mental health status included: "Is your current mental health tied to fears/worries about… [your] past cancer and treatment?" Participants were asked to endorse "yes" or "no" in response to this item. An example item regarding the perceived impact of COVID-19 on daily living included: "What COVID-19 restrictions are currently in place where you live?" Participants were asked to select all that applied to them from a series of options, such as "school cancelled," "public gatherings limited to <5 people," and/or "must wear mask/face covering in public." See **Data Sheet 1** for a full version of this COVID-19 questionnaire.

COVID-19-Related Information Needs

Participants were asked to answer 5 questions regarding their COVID-19-related information needs. These items were also developed by the research team and in collaboration with patient partners. Example items included: "Have you received information related to the potential risks of COVID-19 as a survivor of childhood cancer?" and "Would you like to receive more information about your specific risks from COVID-19 as a survivor of childhood cancer?" Participants were also asked to identify specific materials or resources that may help to improve their mental health by selecting all that applied to them from a list of options, such as "information specific to cancer survivors regarding mental health," "online social connections," and/or "general information regarding mental health." See **Data Sheet 1** for a full version of this COVID-19 questionnaire.

Demographic Information

Participants completed a demographic form. Information regarding their date of birth, sex, gender, ethnicity, and country of residence was collected.

Cancer History

Participants answered questions regarding their cancer history, including their age of diagnosis, cancer diagnosis, type of treatment, and years of treatment. Three items adapted from the Self-Report Survey of Cancer Knowledge by Kunin-Batson et al. (2016) were also included. An example item is: "Did your cancer treatment cause any health problems you are currently experiencing?"

Statistical Analyses

Preliminary screening was conducted to assess for any missing data, outliers, multicollinearity, and normality. All analyses were computed on SPSS 27.0 (IBM Corp, 2020). To address Aim 1, we provided a summary of descriptive statistics of participant responses, including their clinical characteristics and mental health status. Pairwise deletion was used to handle missingness in descriptive analyses. For exploratory purposes, we conducted independent *t*-tests and a one-way ANOVA to explore differences in patient and clinical characteristics in relation to study outcomes. We conducted independent *t*-tests to explore how differences between participants that reported their current mental health status was tied to fears/worries related to their past cancer and treatment, with those who denied the same, in relation to their mental health status, indexed by anxiety, depression, and PTSS. PTSS scores were standardized to combine data collected from CPSS-5 and PCL-5. All *t*-tests were bootstrapped to generate confidence intervals (CIs) and further reduce any effects of possible non-normality or outliers. Where possible, effect sizes (Cohen's *d;* Cohen, 2013) are reported. To address Aim 2, we provided a summary of descriptive statistics of participant responses on their report of their COVID-19-related information needs. We also developed and disseminated an infographic summarizing preliminary findings and evidence-based, recommended strategies for coping with the pandemic.

RESULTS

Participant and Clinical Characteristics

One hundred and six participants originally completed the study questionnaire. However, 12 participants were removed due to ineligibility as per study criteria (8 participants were over 39 years old, 1 participant mis-entered their date of birth, 3 participants did not report their date of birth). As a result, a total of 94 participants were included in the data analysis with a mean age of 26.9 years (SD = 6.2). Of this sample, 79 participants (84.0%) reported that their sex assigned at birth was female, 14 participants reported that sex assigned at birth was male, and 1 participant preferred not to answer. Regarding their gender, 78 participants identified as female, 13 as male, 2 as non-binary, and 1 preferred not to answer. Participants were asked to describe their ethnicity among listed categories and indicate all that applied to them. They most commonly identified as White ($n = 88$; 93.6%), multi-ethnic ($n = 9$; 9.6%), and East Asian ($n = 4$; 4.3%). Participants reported residing in 10 countries, most commonly Canada ($n = 37$; 40.2%), the United States ($n = 28$; 30.4%), and the UK ($n = 6$; 15.2%).

Participants elected to complete questions regarding their cancer history. AYA cancer diagnoses included lymphoma ($n = 30$; 34.1%), leukemia ($n = 26$; 29.5%), solid tumour ($n = 24$; 27.3%), and brain tumours ($n = 8$; 9.1%). The mean age of diagnosis was 11.01 years ($SD = 5.49$) and the mean years off treatment was 14.90 (SD = 8.35). The types of treatment received included chemotherapy ($n = 84$; 95.5%), surgery ($n = 67$; 76.1%), radiation ($n = 32$; 36.4%), and bone marrow transplant ($n = 7$; 8.0%). A summary of participant and clinical characteristics is provided in **Table 1**.

Differences in participant characteristics, including sex and gender, were explored in relation to study outcome variables, including anxiety, depression, and PTSS. No significant differences emerged. Likewise, differences in type of cancer diagnosis in relation to the same study outcome variables were explored, and no significant relations were observed. See **Supplementary Table 1** for complete results.

COVID-19 Characteristics

The most common COVID-19 restrictions reported by participants in their respective place of residence were physical/social distancing in public ($n = 60$; 63.8%), community re-launch plans ($n = 60$; 63.8%) and remote learning for school/education ($n = 52$; 55.3%). Five participants (6%) reported that they were infected with COVID-19. Among these individuals, 4 participants (5%) reported that they have recovered from symptoms of COVID-19. All COVID-19 characteristics are provided in **Table 2**.

Aim 1a: To Explore the Perceived Impact of COVID-19 on the Mental Health Status AYA Survivors, Including Ratings of Anxiety, Depression, and PTSS

Participants reported on their perceived risk of severe complications from COVID-19 in comparison to their peers who had not had cancer. More than half of the participants (69.1%) indicated that their risk was *somewhat more* to *much more* than their peers. In addition, 23 participants (28.4%) rated their risk as *about the same* as their peers and 2 participants (2.5%) rated their risk as much less than their peers. These findings are summarized under **Figure 1**.

Participants described their mental health status now compared to prior to the pandemic. Almost half of the participants (49%) rated their mental health status ranged from *somewhat worse* to *much worse* now, while 37 participants (39.4%) rated that their mental health status as having *stayed about the same*. Four participants (4.9%) reported that their mental health status ranged from *somewhat better* to *much better* now. Participants were also asked to describe how well their mental health fared in comparison with their family and friends, with 35 participants (37.2%) reporting that they were *worse*, 37 participants (39.4%%) reporting that they were *about the same*, and 9 participants (10%) reporting they were *better* than those of their family and friends. These findings are summarized under **Figure 1**.

Participants further reported on their mental health status using clinical rating scales of anxiety, depression, and PTSS. While participant scores for anxiety were on average in the *mild* range ($M = 58.8$, SD = 9.3), 33 participants (42.3%) reported

TABLE 1 | Participant and clinical characteristics.

	n (%)	*M* (SD)
Participant Demographic (*n* = 94)		
Age		26.87 (6.23)
Sex		
Female	79 (84.0)	
Male	14 (14.9)	
Other	1 (1.1)	
Gender		
Female	78 (83.0)	
Male	13 (13.8)	
Non-binary	2 (2.1)	
Prefer not to answer	1 (1.1)	
Ethnicity[1]		
White	88 (93.6)	
Identified as multi-ethnic	9 (9.6)	
East Asian (e.g., Chinese, Japanese, Korean)	4 (4.3)	
Arab	2 (2.1)	
Black	2 (2.1)	
South Asian (e.g., East Indian, Pakistani, Sri Lankan)	2 (2.1)	
Aboriginal (First Nations, Inuit, Métis)	1 (1.1)	
Latin American	1 (1.1)	
Southeast Asian (e.g., Vietnamese, Cambodian, Thai)	1 (1.1)	
West Asian (e.g., Iranian, Afghan)	1 (1.1)	
Other	1 (1.1)	
Prefer not to answer	1 (1.1)	
Country of Residence[2]		
Canada	37 (40.2)	
USA	28 (30.4)	
England (Identified separately)	8 (8.7)	
Ireland	7 (7.6)	
UK	6 (6.5)	
Austria	2 (2.2)	
Australia	1 (1.1)	
Finland	1 (1.1)	
Germany	1 (1.1)	
Japan	1 (1.1)	
Clinical Characteristics (*n* = 88)		
Age of Diagnosis (years)		11.01 (5.49)
Cancer Diagnosis		
Lymphoma (e.g., Hodgkin's, non-Hodgkin's)	30 (34.1)	
Leukemia (e.g., ALL, AML)	26 (29.5)	
Solid Tumor (e.g., Wilms' tumor, osteosarcoma)	24 (27.3)	
Brain Tumor (e.g., Medulloblastoma)	8 (9.1)	
Treatment Received[1]		
Chemotherapy	84 (95.5)	
Surgery	67 (76.1)	
Radiation Therapy	32 (36.4)	
Bone Marrow Transplant	7 (8.0)	
Years off-treatment		14.90 (8.35)
Have you had a relapse or second cancer diagnosis?		
No	78 (88.6)	
Yes	10 (11.4)	
Did cancer treatment cause any health problems you are currently experiencing?		
No	32 (36.4)	
Yes	56 (63.6)	
Do you have any other health conditions/concerns?		
No	55 (62.5)	
Yes	33 (37.5)	
Do you feel that your cancer treatment could cause serious future problems?		
No	12 (13.6)	
Yes	59 (67.0)	
I do not know	17 (19.3)	

[1] *Participants were asked to select all that applied. Total number of responses can exceed total number of participants.*

[2] *Two participants did not complete this item (n = 92).*

TABLE 2 | COVID-19 characteristics.

	***n* (%)**
What COVID-19 restrictions are currently in place where you live?	
Community beginning to re-open stores and services	60 (63.8)
Must maintain physical or social distance when in public	60 (63.8)
School being offered *via* remote learning	52 (55.3)
School cancelled	40 (42.6)
Must wear mask/face covering in public	29 (30.9)
Public gatherings limited to <5 people	23 (24.5)
Public gatherings limited to <50 people	21 (22.3)
Stay at home order (except for essential work or outings)	18 (19.1)
Public gatherings limited to <15 people	15 (16.0)
Others	7 (7.4)
Curfew	3 (3.2)
No restrictions	3 (3.2)
Have you been told by a doctor or other health care professional that you have, or have had COVID-19?	
No	76 (93.8)
Yes, and the condition is no longer present	4 (4.9)
Yes, and the condition is still present	1 (1.2)
Have you been exposed to someone who has been diagnosed with COVID-19	
No	74 (91.4)
Yes	7 (8.6)

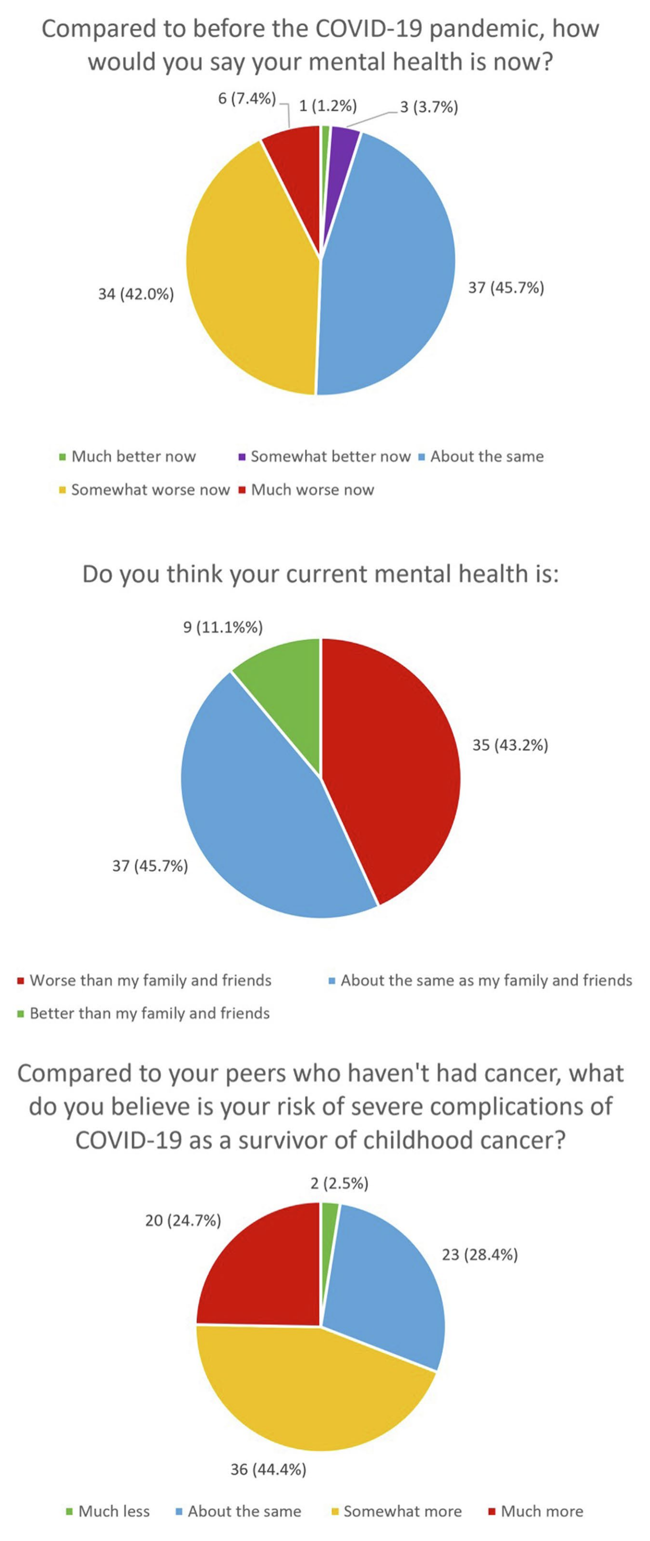


FIGURE 1 | The perceived impact of COVID-19 on the mental health status of AYA survivors.

experiencing *moderate* to *severe* levels of anxiety. Additionally, participant scores for depression were on average in the *mild* range (*M* = 55.4, SD = 10.3), and 22 participants (28.2%) reported experiencing *moderate* to *severe* levels of depression. Finally, participant scores of PTSS for adolescent survivors that completed the CPSS were on average below the clinical cutoff (*M* = 28.3, SD = 20.6) with one participant (1.1%) reporting clinically significant levels of PTSD symptoms. Scores of PTSS for young adult survivors who completed the PCL-5 were on average below the clinical cutoff (*M* = 22.0, SD = 18.0) with 18 participants (19.1%) indicating clinically significant levels of PTSD symptoms.

Participants identified factors that impacted their mental health status during COVID-19. Notably, 39 participants (41.5%) indicated that their current mental health status was tied to fears/worries about their past cancer. Similarly, 40.4% (*n* = 38) of respondents indicated that their mental health status was related to fears/worries about catching COVID-19. The remainder of participants identified media and messaging about the pandemic (19.1%, *n* = 18), nothing in particular (10.6%, *n* = 10), other (5.3%, *n* = 5; e.g., "no supports available with new baby"), and not applicable (2.1%, *n* = 2) as factors related to their mental health status.

Aim 1b: To Compare the Mental Health Status of AYA Survivors Who Reported That Their Mental Health Status Was Tied to Fears/Worries About Their Past Cancer and Treatment, With Those Who Did Not Report the Same Experience

Thirty-eight participants who indicated that their current mental health status was tied to fears/worries about their past cancer and treatment, reported significantly higher levels of anxiety (*M* = 62.05, SD = 8.15; *moderate* range) compared to the 40 participants who did not report the same (*M* = 55.71, SD = 9.33; *mild* range; *t*(76) = 3.19, *p* = 0.002, *d* = 0.72, 95% CI [2.38, 10.30]). No significant difference in levels of depression was observed between the 38 participants who shared that their current mental health status was tied to fears/worries about their past cancer history ($M = 57.17$, SD = 10.25; *mild* range) compared to the 40 participants who did not report the same association ($M = 53.67$, SD = 10.25; *none to slight* range; $t(76) = 1.51$, $p = 0.135$, $d = 0.34$, 95% CI [−1.12, 8.13]). Finally, 37 participants who endorsed their current mental health status was tied to fears/worries about their past cancer and treatment ($M = 0.34$, SD = 1.07) reported significantly higher levels of PTSS than the 38 participants who did not endorse the same ($M = -0.33$, SD = 0.79, $t(73) = -3.12$, $p = 0.003$; $d = 0.94$, 95% CI [−1.11, −0.24]). These results are reported in **Table 3**.

Aim 2a: To Describe the COVID-19-Related Information Needs of AYA Survivors

Over three-quarters of the participants (76.5%) indicated that they had not received any information related to the potential risks of COVID-19 as a survivor of childhood cancer, while 19.8% reported they had received some information, and 3.7% indicated that they did not know. Of note, 13.8% of participants did not answer this question. Of those who had received information related to potential risks of COVID-19, 18.8% reported it had been *definitely* helpful, 50.0% as *moderately* helpful, and 31.3% as *slightly* helpful. Participants were also asked if they would like to receive more information regarding risks and guidelines and 69.1% endorsed that they would like to receive more information on their specific risks from COVID-19 as a survivor of childhood cancer, while 59.3% of participants wanted more information about guidelines and recommendations for survivors of childhood cancer during the COVID-19 pandemic. These data are presented in **Table 4**.

Aim 2b: To Develop and Disseminate an Infographic Detailing Evidence-Based Coping Strategies for COVID-19 Specific to Survivors of Childhood Cancer

Based on the outcomes identified above, our research team developed and disseminated an infographic summarizing preliminary findings from the current study, along with recommended strategies for coping with their mental health in the pandemic. Content development was informed by our research team through expert consultation. Illustrations were created by graphic designers that specialize in scientific illustrations (Scientific Illustrations, 2022). Feedback of initial iterations was gathered from the research team and patient partners. This iterative process resulted in the development of an illustration focused on: (1) the purpose of the research, participant characteristics, timing of the study and study location, as well as the study rationale; (2) the mental health status of survivors, and factors that impacted their mental health; (3) concerns expressed by survivors regarding their health risks from COVID-19 identified through the current study; (4) recommended coping strategies (e.g., diet, sleep, exercise) and access to online (e.g., Center for Disease Control and Prevention; CDCBreastCancer, 2022) and local, Canadian

TABLE 3 | Independent *t*-tests comparing the beliefs of AYA survivors of childhood cancer regarding their current mental health in relation to their past cancer treatment and experience (2: fears/worries, no fears/worries) and how this related to their current mental health (3, anxiety, depression, PTSS).

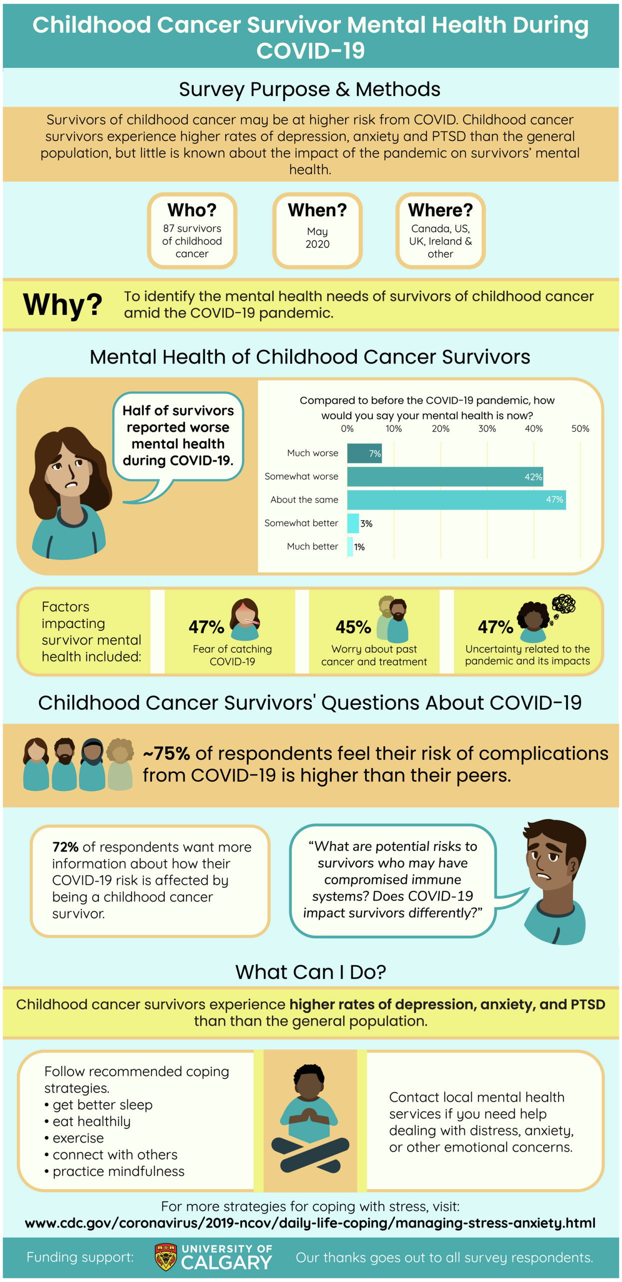
	Fears/Worries			No Fears/Worries								
	n	***M***	***SD***	***n***	***M***	***SD***	***t*-test**	***df***	***p***	**Cohen's *d***	**95%**	**CI**
Anxiety	38	62.05	8.15	40	55.71	9.33	3.19*	76	0.002	0.72	2.38	10.30
Depression	38	57.17	10.25	40	53.67	10.25	1.51	76	0.135	0.34	−1.12	8.13
PTSS†	37	0.34	1.07	38	−0.33	0.79	−3.12*	73	0.003	0.94	−1.11	−0.024

**$p < 0.001$. †PTSS was computed using the CPSS for survivors < 18 years and the PCL-5 for survivors ≥ 18 years. Scores were standardized to compute a combined index of PTSS.*

TABLE 4 | COVID-19-related information needs of AYA survivors.

	***n* (%)**
Have you received information related to the potential risks of COVID-19 as a survivor of childhood cancer	
No	62 (76.5)
Yes	16 (19.8)
I do not know	3 (3.7)
Where did you receive this information?	
My healthcare team	11 (11.7)
Childhood cancer specialist organizations	9 (9.6)
Family and/or friends	2 (2.1)
Mass media	0 (0.0)
Social media	0 (0.0)
Other	0 (0.0)
This information has been:	
Definitely helpful	3 (18.8)
Moderately helpful	8 (50.0)
Slightly helpful	5 (31.3)
Would you like to receive more information about your specific risks from COVID-19 as a survivor of childhood cancer?	
No	25 (30.9)
Yes	56 (69.1)
Are there any materials or resources that may help improve your mental health at this time?	
Information specific to cancer survivors regarding mental health	34 (36.2)
Not applicable	30 (31.9)
Online social connection with other survivors	18 (19.1)
General information regarding mental health	15 (16.0)
Online connection with health-care providers	13 (13.8)
Other	2 (2.1)
Would you like to receive more information about guidelines and recommendations for survivors of childhood cancer during the COVID-19 pandemic?	
No, I do not need specific recommendations for survivors	33 (40.7)
Yes, I would like specific recommendations	48 (59.3)

health services for help. The infographic was shared widely through our research team, patient advocacy groups and community organizations (e.g., Kids Cancer Care Foundation), and social media (i.e., Twitter) in March 2021. As of January 2022, a review of Twitter analytics showed a total of 11, 903 impressions, 502 engagements, 56 "likes" and 22 re-shares (or "retweets") of the infographic disseminated. The infographic is displayed in **Figure 2**.



Childhood Cancer Survivor Mental Health During COVID-19

Survey Purpose & Methods

Survivors of childhood cancer may be at higher risk from COVID. Childhood cancer survivors experience higher rates of depression, anxiety and PTSD than the general population, but little is known about the impact of the pandemic on survivors' mental health.

Who? 87 survivors of childhood cancer

When? May 2020

Where? Canada, US, UK, Ireland & other

Why? To identify the mental health needs of survivors of childhood cancer amid the COVID-19 pandemic.

Mental Health of Childhood Cancer Survivors

Half of survivors reported worse mental health during COVID-19.

Factors impacting survivor mental health included:

47% Fear of catching COVID-19

45% Worry about past cancer and treatment

47% Uncertainty related to the pandemic and its impacts

Childhood Cancer Survivors' Questions About COVID-19

~75% of respondents feel their risk of complications from COVID-19 is higher than their peers.

72% of respondents want more information about how their COVID-19 risk is affected by being a childhood cancer survivor.

"What are potential risks to survivors who may have compromised immune systems? Does COVID-19 impact survivors differently?"

What Can I Do?

Childhood cancer survivors experience **higher rates of depression, anxiety, and PTSD** than than the general population.

Follow recommended coping strategies.
- get better sleep
- eat healthily
- exercise
- connect with others
- practice mindfulness

Contact local mental health services if you need help dealing with distress, anxiety, or other emotional concerns.

For more strategies for coping with stress, visit:
www.cdc.gov/coronavirus/2019-ncov/daily-life-coping/managing-stress-anxiety.html

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FIGURE 2 | Infographic for AYA survivors of childhood cancer on coping with COVID-19.

DISCUSSION

This study explored the perceived impact of the COVID-19 pandemic on the mental health status of AYA survivors. Of the 40% of respondents reporting that their mental health status amid the early stages of the COVID-19 pandemic was linked to their fears/worries about their past cancer and treatment, they reported experiencing greater levels of anxiety and PTSS than those that did not report the same. Additionally, AYA survivors perceived themselves to be at a higher risk from COVID-19 than their family and friends. Despite this, less than a quarter of the participants had received information related to their potential risk, and over half of the participants expressed an interest in receiving more health information. This finding led to our development and dissemination of an infographic reporting our research process and evidence-based, tailored recommendations for how AYA survivors can cope with their mental health during the pandemic.

Nearly half of the participants described their mental health status as worse now compared to before the pandemic. These results are concerning as it is well documented that survivors of childhood cancer already are more likely to experience mental health issues compared to their peers that did not have cancer (Forster and Schulte, 2021; National Cancer Institution, 2022). This means that, for many, they may have already been grappling with anxiety, depression, and PTSS prior to the pandemic and that the onset of COVID-19 only added a greater difficulty to their struggles, resulting in worsened mental health. Further, we know that there has been a significant burden of mental health difficulties among young people as a result of the pandemic (Lee et al., 2020; Stroud and Gutman, 2021). Our findings contribute to this literature, showing that many AYA survivors perceived experiencing poor mental health shortly after the onset of the pandemic and highlighting a need to support this already vulnerable population.

Not all AYA survivors experienced poor mental health during the pandemic. In fact, nearly half of the participants described their mental health status as about the same now as compared to before the pandemic. Further, the same proportion of participants did not express a desire for more information on how to cope with their mental health in the pandemic. The lack of an overwhelming report of the negative impact of COVID-19 may reflect a unique advantage for AYA survivors. For some, their cancer experience may serve as a buffer against some of the risks of the pandemic. A qualitative study conducted by Shay et al. (2021) with AYA patients and survivors identified perceived unexpected advantage of a cancer history during the early stages of the global pandemic. Specifically, AYA survivors relied on strategies that they previously used to cope with their cancer and treatment. Many also accessed social support through online cancer-specific networks that they previously established during their cancer experience (Schulte et al., 2020). Other, more recent studies have found a similar sign of resilience and protective mechanisms identified in survivors and their families as they navigate the pandemic (e.g., Wimberly et al., 2021; Jacobson et al., 2022). In effect, not all AYA survivors are struggling with their mental health due to their past cancer and treatment. Rather, there may be existing areas of strength and/or skills that AYA survivors can leverage for their coping to help alleviate or buffer some of the consequences of living with COVID-19. These discoveries can inform future research priorities on developing interventions for AYA survivors in coping with their mental health during this on-going pandemic.

AYA survivors who reported that their mental health status was tied to fears/worries about their past cancer and treatment reported significantly worse anxiety and PTSS than those who did not report the same. This, along with an expressed interest in receiving more health information, suggests that AYA survivors may benefit from learning more about their potential COVID-19-related health risks and the way in which this information is delivered should account for these perceived mental health vulnerabilities. However, there appears to be a knowledge gap among survivors of childhood cancer in receiving information related to their specific risks both before and during the pandemic. In fact, only 35% of survivors recognize that they could develop a serious health problem as a result of their cancer history (Kadan-Lottick et al., 2002). As health care providers and researchers, it is a priority that we find better ways to share knowledge with this population.

This study provided the basis for the development and dissemination of an infographic as a means to share health information with AYA survivors, including evidence-based recommendations for mental health coping strategies. The creation of this infographic in collaboration with our patient partners was intended to ensure the engagement and input of those with lived experience as an AYA survivor of pediatric cancer in the research process (Staniszewska et al., 2017). We communicated the nature of our research project to AYA survivors with the goal of increasing transparency and accessibility of our work. Further, in the context of a quickly changing global pandemic, we produced this infographic in an effort to quickly respond to the acute needs of AYA survivors early in the pandemic and in turn support their on-going coping with their mental health and overall quality of life.

It is worth noting that current methods of disseminating health information to survivors (e.g., survivorship care plans) are not always effective (Jacobsen et al., 2018). A recent meta-analysis showed that survivorship care plans do not improve patient-reported outcomes, including anxiety, depression, or other cancer-related distress (Hill et al., 2020). Likely, a multi-pronged approach is required to reach survivors, partnering with patients on how and what to distribute are important considerations. There are studies that show AYA survivors need a better way to learn about and engage in their own health information, including meeting them where they are, increased accessibility, and greater equity in receipt of care (Oeffinger et al., 1998; Mertens et al., 2004; Taylor et al., 2004). The development of innovative, childhood cancer-specific, and personalized interventions are just beginning (Devine et al., 2018; McLoone et al., 2020). Our development of an infographic is a unique contribution to this line of inquiry, and future research is needed to evaluate the effectiveness of disseminating these knowledge translation tools, as well as the feasibility of implementation.

There are several limitations to be addressed in future research. The effects of the COVID-19 pandemic are inherently complex. We only examined the mental health status of AYA survivors, as indexed by anxiety, depression, and PTSS at the individual level. Other, multi-level factors were not accounted for. For example, due to insufficient sample size, the current research does not account for group (e.g., country) level analysis that may underlie the current findings (e.g., Lai et al., 2020). Additional studies that incorporate multi-level techniques with adequate sampling distribution may help to clarify some of the influences at the societal and global level on the well-being of AYA survivors. Further, we relied on self-report from AYA survivors regarding their perception of their potential risks to COVID-19. We did not capture open-ended responses to seek specific reasons or motivations behind AYA survivors' perception on this matter. Future research capturing these insights through open-ended response format or qualitative interventions may help to contextualize the experiences of AYA survivors. Our cross-sectional assessment of AYA survivors only allowed us to examine the study participants at one point in time. This did not allow us to capture the possible impact of distinct COVID-19 waves on the experiences of AYA survivors over time. These potential impacts of COVID-19 are dynamic and chronic, as are the needs of AYA survivors. Future studies would benefit from a longitudinal evaluation of the experiences of AYA survivors over time in order to further examine the interactional nature of their experiences coupled with their development level, as well as with the pandemic across time. Inclusion of a data aggregation tool on the COVID-19 trajectory may help to address these possibilities and capture changes over time. In addition, there was potentially a sampling bias inherent in our recruitment strategy that may have hindered our efforts in ensuring a representative and diverse sample of participants. Moreover, there was a lack of representation in our sample of AYA survivors collected. This limited our ability to generalize findings from the current research to diverse groups. Research suggests that individuals who face systematic health and social inequities are at a greater risk of getting sick and dying (Dalton et al., 2008; Bambra et al., 2010; Devine et al., 2018). Therefore, future research ensuring a representative group, such as an inclusion of those who are non-English speaking, from different migration status, and/or those from rural geographic regions, will be essential to ensuring we are capturing the lived experiences of all individuals.

CONCLUSION

COVID-19 is an on-going and global pandemic with serious implications on the mental health of all individuals. As an at-risk group, AYA survivors may be especially vulnerable to the psychosocial impact of the COVID-19 pandemic. We found that in the early pandemic, between May and June 2020, AYA survivors perceived that their fears and worries about their past cancer and treatment contributed to their current mental health status (anxiety, PTSS). AYA survivors also identified a desire for COVID-19-related information. We therefore developed an infographic to help AYA survivors to have better access to practical health information to support their coping and quality of living. With the continued emergence of new SARS-CoV2 variants coupled with the widespread distribution of COVID-19 vaccines and treatments, future research focused on a comprehensive and longitudinal assessment of the mental health status of AYA survivors will be helpful to determine ways that we can continue to support the psychosocial needs of this vulnerable population during this unprecedented crisis.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Health Research Ethics Board of Alberta – Cancer Committee (HREBA.CC-20-0151). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

FS contributed to study conceptualization, methods, data collection, knowledge translation/dissemination (infographic), and writing (preparing the original draft and reviewing and editing). SH contributed to conducting data analysis and writing (preparing the original draft and reviewing and editing). CF, EA, and MS contributed to study development and data collection. AT and SC contributed to data analysis and writing (preparing the original draft and reviewing and editing). VF, CW, LW, LH, GM, PP, and KR contributed to study conceptualization, knowledge translation/dissemination (infographic, review, and feedback), and writing (reviewing and editing). All authors contributed to the article and approved the submitted version.

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Study data were collected and managed using REDCap electronic data capture tools hosted at the University of Calgary Clinical Research Unit. REDCap (Research Electronic Data Capture) is a secure, web-based application designed to support data capture for research studies, providing: (1) an intuitive interface for validated data entry; (2) audit trails for tracking data manipulation and export procedures; (3) automated export procedures for seamless data downloads to common statistical packages; and (4) procedures for importing data from external sources. Illustrations were created by Designs that Cell, graphic designers that specialize in scientific illustrations.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2022.867151/full#supplementary-material

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Adolescent and Young Adult Initiated Discussions of Advance Care Planning: Family Member, Friend and Health Care Provider Perspectives

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Background and Aims: End-of-life (EoL) discussions can be difficult for seriously ill adolescents and young adults (AYAs). Researchers aimed to determine whether completing *Voicing My CHOiCES* (VMC)—a research-informed advance care planning (ACP) guide—increased communication with family, friends, or health care providers (HCPs), and to evaluate the experience of those with whom VMC was shared.

Methods: Family, friends, or HCPs who the AYAs had shared their completed VMC with were administered structured interviews to assess their perception of the ACP discussion, changes in their relationship, conversation quality, and whether the discussion prompted changes in care. Open-ended responses underwent thematic analysis.

Results: One-month post-completion, 65.1% of AYA had shared VMC completion with a family member, 22.6% with a friend, and 8.9% with an HCP. Among a sample of respondents, family (47%) and friends (33%) reported a positive change in their relationship with the AYA. Participant descriptions of the experience fell into five themes: positive experience (47%), difficult experience (44%), appreciated a guide to facilitate discussion (35%), provided relief (21%), and created worry/anxiety (9%). Only 1 HCP noted a treatment change. Family (76%), friends (67%), and HCP (50%) did not think the AYA would have discussed EoL preferences without completing VMC.

Conclusions: VMC has potential to enhance communication about ACP between AYA and their family and friends, though less frequently with HCPs. Participants reported a positive change in their relationship with the AYA after discussing VMC, and described experiencing the conversation as favorable, even when also emotionally difficult.

Keywords: AYA family, friends, HCP, adolescent and young adult, advance care planning, EoL discussions, Voicing My CHOiCES, communication

INTRODUCTION

The number of AYAs living with serious illnesses such as cancer is growing globally (Viner et al., 2011; Cohen and Patel, 2014; GBD 2019 Adolescent Young Adult Cancer Collaborators, 2022). In 2020, an estimated 90,000 adolescents and young adults (AYA) were diagnosed with cancer in the United States (Haines et al., 2021). For many, death is an inevitable outcome of their disease, making advance care planning (ACP) a critical component of care (Brown and Sourkes, 2006; DeCourcey et al., 2021). It is difficult for AYAs, their families, and providers to think about or talk about death and dying. The presence of a life-threatening illness adds a multitude of challenges to what is already a difficult period of life, when emerging adults strive to define themself outside of the context of their family and envision their own future (Brown and Sourkes, 2006). Further, disease burden at this age can negatively impact financial security, body image, educational and work trajectories, relationships with spouse/significant other, and plans for having children (Maslow et al., 2011; Warner et al., 2016; Jin et al., 2017). In addition, young adults with advanced cancer have reported significant psychological distress in the form of grief (Jacobsen et al., 2010) and suicidal ideation (Walker et al., 2008).

Effective end-of-life (EoL) discussions are critical for AYAs, especially in the event of disease progression or a poor prognosis, given both the medical challenges and psychosocial risk factors involved (Sansom-Daly et al., 2020). ACP documents and advance directives provide patients with the opportunity to express their preferences for care. These directives can help families and health care agents make informed decisions, alleviate distress (Mack et al., 2005), avoid decisional regret (DeCourcey et al., 2019; Lichtenthal et al., 2020), and potentially improve the patient's quality of life by respecting their religious, cultural, and familial values and beliefs (Jankovic et al., 2008; Barfield et al., 2010; Kane et al., 2011; Wiener et al., 2012). Families have expressed significant interest in ACP, with parents indicating that the opportunity for these discussions has been a poorly met need (Durall et al., 2012; Lotz et al., 2013; DeCourcey et al., 2019; Hein et al., 2020; Orkin et al., 2020). Research has shown that parents of seriously ill children desire earlier, ongoing opportunities to address ACP with their child's providers (DeCourcey et al., 2019; Orkin et al., 2020). However, many pediatric providers report a lack of ACP communication training (Dellon et al., 2010; Durall et al., 2012; Lotz et al., 2013; Heckford and Beringer, 2014). Unfortunately, when ACP discussions do take place, they often occur too late and typically during an acute clinical crisis, when there is insufficient time to consider individual goals and values (Davidson et al., 2007; Brudney, 2009; Durall et al., 2012; Snaman et al., 2020; Pennarola et al., 2021).

The literature has clearly demonstrated the need and desire for ACP intervention in this population (Weaver et al., 2015; Kirch et al., 2016), as well as the barriers to initiating ACP (Wolfe et al., 2008; Smith et al., 2012; Kassam et al., 2013; Pinkerton et al., 2018). Studies have shown that less than 3% of AYAs participate in EoL planning conversations without clinician prompting (Lyon et al., 2004, 2014; Liberman et al., 2014; Carr et al., 2021). Data exploring the experience of the family members, friends, and health care providers who are involved in ACP conversations with AYA are limited. Adolescents and families who participated in family-centered ACP found the conversations to be worthwhile (Dallas et al., 2016), notably with a greater understanding of EoL wishes (Madrigal et al., 2017). In a robust multi-site, assessor-blinded, parallel-group, randomized control trial (*FACE pACP*), ACP surrogates were eight times more likely than controls to have an excellent understanding of adolescent patients' treatment preferences (Lyon et al., 2018). In another ACP trial, families had more positive appraisals of their caregiving, than families who did not have these conversations (Baker et al., 2020).

Novel tools and interventions are needed to facilitate ACP discussions between AYA and their family members, friends and HCPs (Snaman et al., 2020). *Voicing My CHOiCES* (VMC), a research informed ACP guide (Wiener et al., 2012; Zadeh et al., 2015), has been shown to both decrease anxiety around EoL planning and enhance communication with both family members and friends (Wiener et al., 2021). This study adds to the literature by providing the perspectives or outcomes on behalf of the family member, friend, or HCP post completion of an ACP document by an AYA. In this study we aimed to gain understanding of the experience of the family member, friend, or HCP pertaining to the ACP discussion, changes in their relationship, conversation quality, and whether the discussion prompted changes in care.

MATERIALS AND METHODS

Study Recruitment and Enrollment

As part of a larger study, AYAs aged 18–39 years receiving cancer-directed therapy or treatment for another chronic medical illness at one of seven study sites were enrolled on a larger study examining psychosocial outcomes after completing VMC. For this sub-study, participants included the family members, friends, or HCPs with whom the AYAs initiated a conversation with about their ACP preferences following completion of the VMC guide. Sub-study participants were contacted by phone, with permission from the AYA by whom they had been nominated. The NIH Institutional Review Board approved this protocol, and the study was then approved by the IRB at each of the participating sites. Data was collected between 2015–2019.

Study Procedures

AYAs were contacted one-month post-VMC completion to seek permission to contact any family member, friend, or HCP the AYA had shared preferences with. Informed consent was subsequently obtained and each participant was then administered a one-time structured interview, including both quantitative and open-ended questions. Development of the interview was based on shared clinical expertise of the primary study team, familiarity with the VMC guide, and knowledge of the relevant literature and gaps therein (Wiener et al., 2012, 2021; Dallas et al., 2016; Sansom-Daly et al., 2020). Specifically, the interview assessed the communication they had about ACP with the AYA, as well as perceived changes in their relationship, the quality of the conversation, and whether changes in care were made following the discussion. Interviews were conducted either in person or by phone and responses were written verbatim by the interviewer. No audio or video recordings were collected. Each interview took approximately 15 min to complete. See **Supplemental Data Sheet 1** for the interview guide.

Interviews were conducted by a trained study team member, including psychologists, social workers, nursing study coordinators, or graduate students. Procedure training consisted of an in-person, virtual or phone session with the sponsor site (SZB or LW) where a training manual was reviewed in detail and sample case scenarios were discussed.

Analysis

Responses to open-ended questions were analyzed using a realist approach to inductive thematic analysis (Braun and Clarke, 2006; Maxwell, 2012). Coders (SZB, AF, LW) independently read and re-read the data, identifying initial codes, capturing novel content, and searching for potential themes (Braun and Clarke, 2006; Miles et al., 2014). The coders then met as a group to review codes, and to examine, refine and define themes (Macqueen et al., 1998). Discrepancies were resolved through consensus discussion. Free-text responses were then coded in parallel (Malterud, 2001). The authors reviewed and discussed the findings and summarized the data.

RESULTS

One month after completing the baseline measure and reviewing the VMC tool, 129 participants answered the follow up questions about talking with family members, 124 about talking with friends, and 124 answered the question about talking with HCP. Overall, 84 (65.1%) of participants had shared what they wrote in VMC with a family member and 11 (8.9%) shared with an HCP (Wiener et al., 2021). Twenty-eight participants (22.6%) shared what they wrote in VMC with a friend. Of those with whom document completion was shared, we interviewed 40 (47.6%) family members, 6 (21.4%) friends, and 5 (45.5%) providers about their experience with this conversation. Of note, three interviews (two family members and one provider) were discontinued when the participant indicated the AYA had not shared what they had written in VMC. The remaining analyses are based on interviews with the 48 participants who engaged in such a conversation, according to both the AYA and the study participant.

ACP Discussions Had Pre- and Post-VMC

For 17 (42.5%) of the 40 family members interviewed, their first ACP conversation was held post-AYA VMC completion. Of the friends interviewed, 4 of the 6 friends (66.7%) were from AYAs who first spoke to their friend after VMC. Of the five providers interviewed, 3 (60%) were from AYAs who only spoke to their HCP about ACP post-VMC. Twenty-nine (76.3%) family members, 4 friends (67%) and 2 HCP (40%) did not think the AYA would have talked to them about their EoL preferences without the study. When participants were asked *"Can you tell me what part of the advance care planning process [the patient] shared with you,"* four themes were revealed: preferences on comfort/support, care preferences when critically ill, planning for remembrance, and care/concern for others after death. Sample responses are provided in **Table 1**. One HCP (20%) noted a treatment change following the discussion (e.g., medication changes for symptom management).

Changes in Relationship

Forty-seven percent of family members (n = 19) and 33% of friends (n = 2) reported a change in their relationship with the AYA following the discussion. If a change in their relationship was reported, participants were then asked to describe the change. Four themes were found: the conversation opened lines of communication, increased feelings of closeness, learned something that was important, and changed view of character (i.e., how the AYA thinks, feels or copes). Sample responses are provided in **Table 2**.

How the ACP Conversation Was Experienced

Family members, friends and HCPs were also asked, "*Can you tell us what this experience was like for you?*" What emerged illustrated both benefit and burden. Themes that represented benefit included the conversation being a positive experience, appreciating a guide to facilitate a deeply honest conversation and the conversation provided relief. Themes that represented a burden included experiencing the conversation as emotionally difficult and that it created worry/anxiety. While some participants described a sense of burden from the discussion (e.g., difficult experience or created worry/anxiety), the majority described benefit (e.g., positive experience, appreciated guide, provided relief). Of those participants who reported a burdensome experience most indicated finding benefit despite the burden (e.g., painful to have the discussion but grateful to know what their EoL preferences are). Select participant responses are provided in **Table 3**.

DISCUSSION

ACP discussions have been associated with a range of positive outcomes, including increased congruence between treatment preferences expressed by AYAs and their caregivers and increased likelihood that these preferences will be honored at the EoL. Yet, AYAs and their caregivers find it difficult to engage in these conversations (Jimenez et al., 2018). For the majority of study participants, completing an age-appropriate ACP guide prompted a first conversation regarding EoL preferences with a family member. To a lesser degree, it also prompted a first conversation with a friend. Notably, many of these conversations covered more than just EoL preferences. Participants described having deeply honest discussions about hopes, fears, and relationships. These findings support using VMC to enhance communication about EoL preferences, adding to the existing literature on the myriad benefits of such interventions (Feraco et al., 2016; Lin et al., 2020; Laronne et al., 2021).

TABLE 1 | Part of ACP shared with participant post-VMC completion.

Shared component of ACP	Coding definition	Participant responses
Preferences on Comfort/Support	Discussed ways to provide comfort and support, including how to manage visitation	*"Doesn't want a lot of people visiting if very sick."* *"He shared how he wants to be treated when not feeling well and what's comforting to him, even in his room."* *"A list of who could visit, and "no one can cry." She wants mental health professionals on call for visitors."* *"Emotional support if things don't go well. Someone to call or be there for him if he gets really sick"*
Care Preferences When Critically Ill	Discussed who will make care decisions if they are unable to and what kind of care preferences they have, including life support options and where they want to be at the end of life	*"If he would get worse, he would want his older brother to make life support decisions for him. And me too. . .He shared that he did not want his dad to make decisions because his dad would never say no to life support, and he does not want to live on life support unless it is reversible."* *"The chain of command if something happened to him."* *"We went over worst-case scenarios. She read through it with me. It brought up issues we hadn't talked about before. Now, I know what she wants."* *"She said if she was connected to a machine for surviving, she would accept that for a short time but if they found that there was no improvement, and she would not get better she would not be connected to these machines."* *"That he would want to be home at the end."*
Planning for Remembrance	Communicated thoughts on after death and funeral planning, how to distribute/donate belongings	*"What she'd like at a funeral, music, and how she would like to be remembered."* *"Would not want an open casket. Wants a celebration of life."* *"The section "How I wish to be Remembered." The main part that was shared was regarding what he would be leaving behind and to whom he would leave certain things (i.e., his personal belongings)."* *"Details about things she had thought about but hadn't spoken to us about- especially after death (Belongings, celebration, cremation, donate her body, where her ashes should go)"* *"What she wants after she dies. No open casket which surprised me. "If I can't see then, they shouldn't be able to see me" Hospice at home.* *"That she would want her ashes going out to sea"*
Care/Concern for Others After Death	Expressed concern for the care of others left behind after their death	*"The will for what happens after she dies. We talked about how the stuff with the kids was missing. We wanted to be able to say where they go, who takes care of them, how finances get to them."* *"His concern about his little sister."* *"It was very focused on her daughter understandably. I wish she had written more specific details of what to do (with her daughter, what traditions to continue etc.)."*

TABLE 2 | Perceived changes in relationship post-VMC discussion.

Theme	Coding definition	Sample response
Opened lines of communication	Created an opportunity for discussion, broke down barriers	*"[study participation] cracked open that door and allowed him to express things he was probably thinking about since diagnosis and questions about whether he will survive. That barrier has now been broken down and we feel we can probably talk about anything."* (*family member*) *"We were avoiding the questions before- broke down barriers."* (*family member*)
Increased feeling of closeness	Enhanced sense of connection between individuals	*"I feel closer to him."* (*friend*) *"These conversations do bring you closer."* (*family member*)
Learned something that was important	Gained knowledge about AYAs preferences and/or values	*"Few things I didn't know* (*his cross/chain*) *and how important those things are to him."* (*family member*) *"I realize more how much she needs me and relies on me."* (*family member*)
Changed view of character	Changed view of how AYA thinks, feels, copes, behaves	*"I look at her as fearless and with more respect"* (*family member*) *"She grew so much from completing this"* (*family member*)

TABLE 3 | Participant perceptions of the ACP discussion.

Theme	Coding definition	Sample quote
Positive experience	Experience was found to be helpful or beneficial	*"It is really good to know what he wants. allowed me to see where she was coming from, not just as a patient, but also as a person. Created open space for her to discuss difficult topics with me."* (*provider*) *"This was a very intimate conversation, it opened doors for us. It brought trust between us that we can now talk more openly. First time we could do this. It was a true gift to me as his mom, to our relationship, and to our whole family."* (*family member*)
Difficult experience	Experience was burdensome, emotionally, for the participant	*"Very hard. I choked up but had to remember this is not happening now, and we need to talk about all of it."* (*family member*) *"Very intense. I didn't expect to get so sad. It's so much more thorough than previous conversations."* (*Family member*)
Voicing My CHOiCES provided an opportunity for discussion	Benefit of the actual ACP guide was endorsed	*"It was nice for this to be available and not just all on me to remember everything she said. Makes it easier on me. And it provides us a vehicle to expand on her thoughts and preferences if she doesn't respond to this next treatment."* (*family member*) *"The document is great because it provides a medium for the conversation. It is also great that it allows conversations to happen when we aren't in crisis, this makes it easier to talk about these things."* (*family member*)
Provided relief	Participant noted feeling better having had the conversation	*"I was glad he had an opportunity to talk to someone other than me. Handling this better at his home center. Glad he's not holding things in. I am more relaxed that he is less stressed. I never want to think about these issues, but I am so relieved since we are both thinking about "it" and neither knew how to broach the subject."* (*family member*) *"Relieved because we were not talking about what was happening."* (*family member*)
Created worry/anxiety	Experience contributed to psychosocial distress	*"There is a lot of pressure/burden on me to be her #1 and her parents aren't even on the list* (*or aren't prominent on the lists of people*)*. It is kind of stressful to think she needs me that much."* (*friend*) *"A lot on me with the family dynamics– I'll be the one to do a lot. Sad/scary to think about going down that path."* (*Family member*)

In addition to families requiring assistance in navigating these discussions (Kenney et al., 2021), we found few AYAs shared what they wrote in VMC with their HCP. Challenges surrounding ACP conversations with HCPs include provider discomfort and a lack of training and resources (Dellon et al., 2010; Durall et al., 2012; Lotz et al., 2013; Heckford and Beringer, 2014). In fact, most family members and friends, as well as half of the HCP in our study, did not think the AYA would have talked to them about their EoL preferences without having completed VMC. This highlights the need for training on how to introduce ACP more comfortably with AYAs so that goals regarding current and future care can be addressed. While just one of five HCPs noted a treatment change following the discussion, these numbers may reflect the limited number of HCPs who the AYAs engaged in an ACP conversation. This is consistent with current literature suggesting patients and their family members will wait for the topic to be raised by their clinician (Clayton et al., 2005; Brighton and Bristowe, 2016). Research focused on training clinicians and preparing patients and families to engage in high-quality discussions using an age-appropriate ACP guide, like VMC, may help to achieve higher quality EoL care.

The specifics of what the AYA shared following completion of VMC was varied and included their preferences on mechanisms of comfort and support, how aggressively they would like to be treated if there was little to no chance of recovery, planning for how they would like to be remembered, and care/concern for others after death. These conversations were well received and described as being beneficial to both parties, despite being emotionally difficult to initiate. Similar to extant literature (Aldridge et al., 2017; Hein et al., 2018; Weaver et al., 2021), participants in the current study recognized that communicating important EoL care preferences can help prepare for future situations and create a pathway to goal-concordant EoL care. Critical longitudinal data is needed to assess whether communicated preferences were honored after an AYAs death.

Other benefits from these conversations were also reported. Half of family members and a third of their friends reported a change in their relationship with the AYA. Changes were all self-reported to be positive. Additionally, when describing the overall experience of talking about ACP with the AYA, participants again highlighted benefit despite also being seen as burdensome. Many participants spoke to the value of the ACP conversations and the relief in having the discussion despite the stress of thinking about worst-case scenarios. These findings can reassure family members and HCPs of advantages associated with these courageous conversations.

Some limitations are important to note. First, many family members (n = 44, 53.7%), friends (n = 6, 54.5%), and HCP (n = 5, 50%) who AYAs shared their VMC completion with were not interviewed. For some, the AYA wasn't comfortable with having the researcher reach out to them, and for others the family member, friend, or HCP was unreachable or declined participation. Second, interviews were only conducted with English-speaking individuals. Therefore, we do not know if the themes that were identified would be different from the family members, friends and HCP who were not interviewed. Third, we contacted AYA participants 1 month after they completed VMC. We don't know if a conversation occurred with a family member, friend, or HCP about what they wrote in VMC past this point. Fourth, demographic data was not collected on the contact participants, so it is unknown whether one demographic was more represented than another. Last, the perspectives obtained might have been affected by recall bias. Despite these limitations, to the best of our knowledge, this is the first multisite study that describes the unique perspectives of family members, friends, and HCP after discussing ACP preferences with AYA post completion of VMC. The concurrent voices captured here poignantly illustrate the shared sense of burden and benefit AYAs and all those who care for them experience trying to communicate during the complex journey at the end of life.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by NIH Institutional Review Board: National Institutes of Health. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

SB, LW, and MP contributed to the conception and design of the study. SB, LW, JT, PM, CH, and KZ collected the data. SB organized the database and wrote the first draft of the manuscript. MG, SB, and AF performed the statistical analysis. LW wrote sections of the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2022.871042/full#supplementary-material

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Patient reported quality of life in young adults with sarcoma receiving care at a sarcoma center

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Background: Sarcomas are a diverse group of neoplasms that vary greatly in clinical presentation and responsiveness to treatment. Given the differences in the sites of involvement, rarity, and treatment modality, a multidisciplinary approach is required. Previous literature suggests patients with sarcoma suffer from poorer quality of life (QoL) especially physical and functional wellbeing. Adolescent and young adult (AYA) patients are an underrepresented population in cancer research and have differing factors influencing QoL.

Methods: Retrospective analysis of Young Adult patients (age 18–39) enrolled in the Sarcoma Tissue Repository at University of Iowa. QoL was assessed using the self-report FACT-G questionnaire at enrollment and 12 months post-diagnosis; overall scores and the 4 wellbeing subscales (Physical, Emotional, Social, Functional) were calculated. Linear mixed effects models were used to measure the association between the rate of change in FACT-G subscale scores and baseline clinical, comorbidity, and treatment characteristics.

Results: 49 patients were identified. 57.1% of patients had a malignancy involving an extremity. Mean FACT-G scores of overall wellbeing improved from baseline to 12 months (76.4 vs. 85.4, $p < 0.01$). Social and emotional wellbeing did not differ significantly between baseline and 12 months. Physical wellbeing (18.8 vs. 23.9, $p < 0.01$) and functional wellbeing (16.8 vs. 20.0, $p < 0.01$) scores improved from baseline to 12 months. No difference was seen for FACT-G overall scores for age, sex, laterality, marital status, performance status, having children, clinical stage, limb surgery, chemotherapy, or tumor size. A difference was demonstrated in physical wellbeing scores for patients with baseline limitation (ECOG 1-3) compared to those with no baseline limitation (ECOG 0) ($p = 0.03$). A difference was demonstrated in social wellbeing based on anatomical site ($p = 0.02$).

Conclusion: Young adults with sarcoma treated at a tertiary center had improvements in overall reported QoL at 12 months from diagnosis. Overall baseline QoL scores on FACT-G were lower than the general adult population for YA patients with sarcoma but at 12 months became in line with general population norms. The improvements seen merit further investigation to evaluate how these change over the continuum of care. Quality of life changes may be useful outcomes of interest in sarcoma trials.

KEYWORDS

quality of life, sarcoma, young adult, oncology-discipline, FACT-G

Introduction

Though underrepresented in research approximately 89,500 adolescents and young adults (AYA) are diagnosed with cancers in the United States (US) annually (Miller et al., 2020). Sarcomas are among the most common cancers in the AYA age group, age 15–39 as defined by the National Cancer Institute with an incidence rate between 1.3 and 3.6 per 100,000 for soft tissue sarcomas and 0.3–1.6 per 100,000 for bone cancers (Miller et al., 2020). Sarcomas are a diverse group of neoplasms that vary greatly in clinical presentation and responsiveness to therapy (Hui, 2016). Given this medical and scientific complexity, and a heterogenous population in terms of sites of involvement rarity, age groups effected, and treatment modalities, a multidisciplinary approach is required with a focus on patient centered care and patient quality of life (Bottomley, 2002; Soliman et al., 2009; Deshpande et al., 2011; Winnette et al., 2017).

Previous literature suggests patients with sarcoma suffer from poorer quality of life (QoL) especially regarding physical and functional wellbeing (Coens et al., 2015; Hudgens et al., 2017). Similarly poor quality of life outcomes in terms of physical and functional wellbeing were seen for AYA patients with sarcoma in AYA HOPE study (Smith et al., 2019). Age groups are affected differently both in terms of the type of sarcoma they have and how this impacts their lives (van der Graaf et al., 2017). QoL in AYA patients may have more of an impact on studies, jobs, and changes in social relationship during the course of their treatment (Fujii et al., 2019). They may also recognize differences in QoL more reliably than their providers (Kaal et al., 2021).

Recent published reviews acknowledge the paucity of literature on quality of life and psychosocial issues in patients with sarcoma (McDonough et al., 2019). There is even less literature regarding AYA patients with sarcoma. Therefore, this study aims to understand if there is an association with treatment at a tertiary sarcoma center and differences in quality of life for young adults with sarcomas.

Materials and methods

Study design and population

A retrospective analysis was undertaken of young adult (YA) patients with sarcoma age 18–39 in the Sarcoma Tissue Repository (STiR) who had an available enrollment questionnaire or 12-month questionnaire wherein at least one of the subscales was completed. Patients under the age of 18 were not enrolled in the registry as patients under 18 are only asked about enrollment at physician request therefore the standard definition of AYA per NCI was not used (Adolescent and Young Adult Oncology Progress Review Group [AYAOPR]., 2006). Data was obtained from review of the University of Iowa Oncology Registry and Sarcoma Tissue Repository (STiR) established in 1992, as well as electronic medical records. Patients were selected who were enrolled in the Sarcoma Tissue Registry and received care at University of Iowa between 2008 (when FACT-G administration began) and 2021. Only patients who had a baseline/enrollment FACT-G filled out within 6 months of diagnosis were included in the study. The planned focus of the study was short term follow up to next 1 year questionnaire looking at patient, disease, treatment, and QoL data.

Demographics

Eastern Cooperative Oncology Group (ECOG) performance status scores were retrieved from clinical notes if reported and if not reported were assigned by a clinician based on information provided in the chart at baseline. Information present in history, exam, or questions regarding activity were used by clinicians to help assess performance. Clinical stage was abstracted from the patient chart and was assigned using NCCN guidelines for the specified site by clinician review if not initially reported.

Outcomes

QoL was assessed using self-reported Functional Assessment for Cancer Therapy-General (FACT-G) questionnaires at baseline enrollment and 12 months. The FACT-G is a well validated, 27 question, 104 point scale that has four subscales assessing physical wellbeing (PWB, 0–28), functional wellbeing (FWB, 0–28), social/family wellbeing (SWB, 0–28), and emotional wellbeing (EWB, 0–24) (Cella et al., 1993; Victorson et al., 2008). Scores were reported as overall wellbeing and 4 subscales; physical, emotional, social and functional.

TABLE 1 Demographics.

Variable	Level	*N* = 49
Sex	Female	21 (42.9)
	Male	28 (57.1)
Race/Ethnicity	Caucasian	48 (98.0)
	Hispanic	1 (2.0)
Laterality	Left	17 (34.7)
	Not paired	16 (32.7)
	Right	16 (32.7)
Performance status (enrollment)	0	24 (49.0)
	1	18 (36.7)
	2	6 (12.2)
	3	1 (2.0)
Performance status	0	24 (49.0)
	1–3	25 (51.0)
Marital status	Married	20 (42.6)
	Single	27 (57.4)
	Missing	2
Has biological children	No	35 (71.4)
	Yes	14 (28.6)
Clinical stage	1	14 (31.8)
	2	13 (29.5)
	3	6 (13.6)
	4	11 (25.0)
	Missing	5
Clinical stage	1–2	27 (61.4)
	3–4	17 (38.6)
	Missing	5
Location	Abdomen	6 (12.2)
	Head	4 (8.2)
	Lower Ext	22 (44.9)
	Other	1 (2.0)
	Pelvis	6 (12.2)
	Thoracic	4 (8.2)
	Upper extremity	6 (12.2)
Trunk vs. Extremity	Extremity	28 (63.6)
	Trunk	16 (36.4)
	Missing	5
Surgery w/in first year	No	3 (6.1)
	Yes	46 (93.9)
Limb surgery w/in first year	No	23 (46.9)
	Yes	26 (53.1)
Type of limb surgery	Amputation	2 (7.7)
	Limb-Sparing	24 (92.3)
	Missing	23
Radiation w/in first year	No	40 (81.6)
	Yes	9 (18.4)
Chemotherapy/targeted/hormone therapy w/in first year	No	17 (34.7)
	Yes	32 (65.3)

Analysis

Linear mixed effects models were used to estimate the overall change in QoL scores between enrollment and 12-month and measure the association between the rate of change in FACT-G QoL scores and patient (e.g., age, gender, marital status), disease (e.g., stage, grade), and treatment (e.g., biopsy, surgery, chemotherapy, radiation) characteristics. Random effects were included to account for the longitudinally correlated nature of repeated QoL assessments at unequal time spacing between visits with a spatial power correlation structure. All statistical testing was two-sided and assessed for significance at the 5% level using SAS v9.4 (SAS Institute, Cary, NC).

This study was reviewed and approved by the University of Iowa Intuitional Review Board (IRB 202106171).

TABLE 2 FACT-G wellbeing scores.

	Questionnaire		
Covariate	**Enrollment**	**Change (12-month–Enrollment)**	****P*-value**
Physical wellbeing	18.8 (16.6, 21.0)	5.1 (2.2, 8.0)	<**0.01**
Social wellbeing	24.4 (23.0, 25.8)	-0.6 (-2.5, 1.4)	0.56
Emotional wellbeing	17.1 (15.6, 18.6)	0.3 (-1.2, 1.8)	0.66
Functional wellbeing	16.5 (14.6, 18.3)	3.3 (1.1, 5.5)	<**0.01**
Overall wellbeing	76.4 (71.3, 81.4)	9.0 (3.5, 14.5)	<**0.01**

*P-values are from linear mixed effects models. †22 patients had only a baseline questionnaire. 15 patients had both a baseline and 12-month questionnaire. 12 patients had only a 12-month questionnaire. Bold values are statistically significant

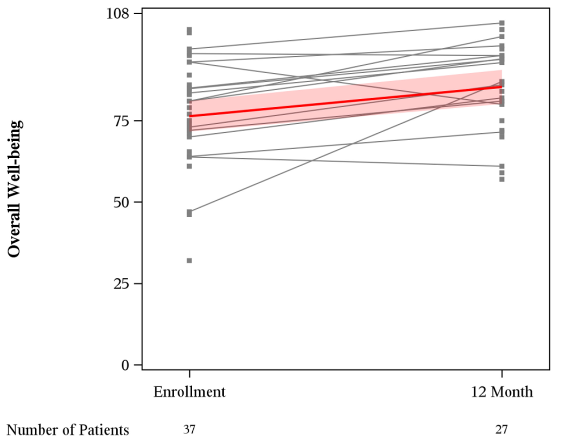

FIGURE 1
FACT-G overall wellbeing score.

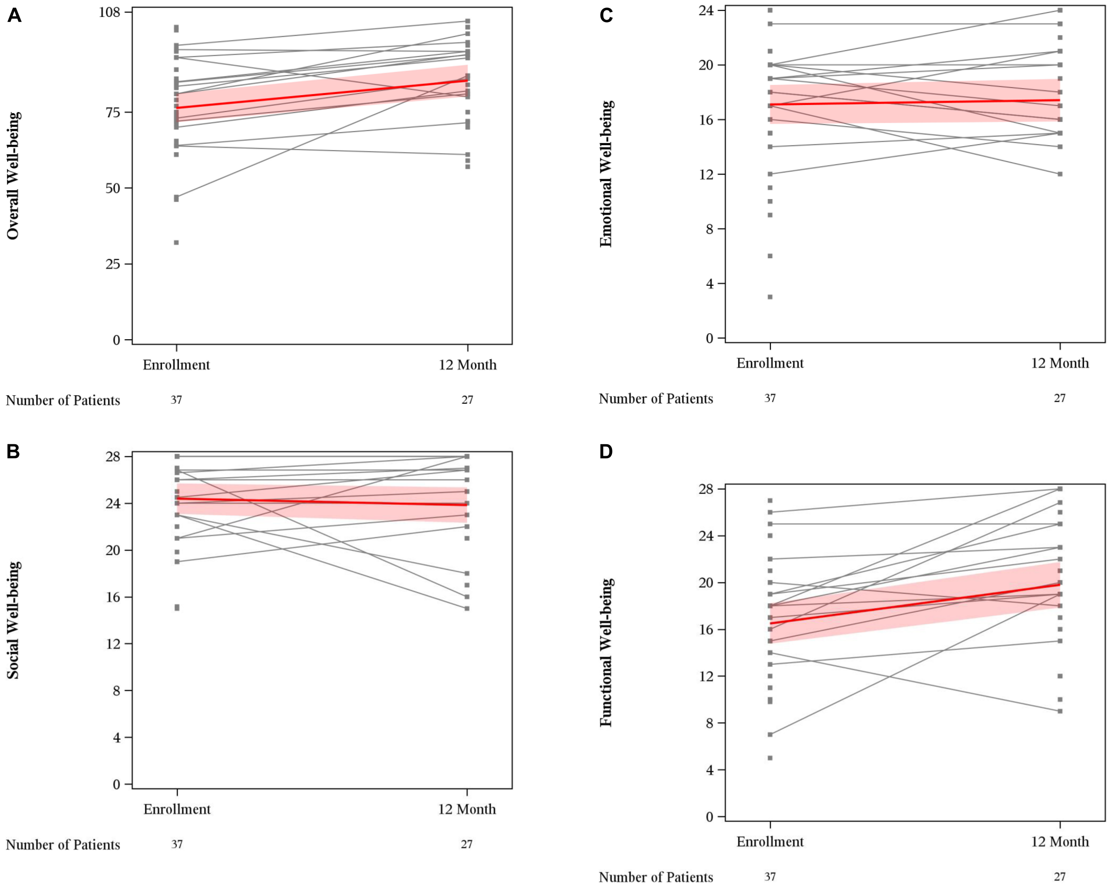

FIGURE 2
FACT G subset scores. **(A)** Overall well-being; **(B)** social well-being; **(C)** emotional well-being; and **(D)** functional well-being.

Results

Demographics

A total of 49 young adult (YA) patients met inclusion criteria for the study. There were 21 (42.9%) female patients. Age at diagnosis had a mean of 29 years [standard deviation (SD) = 6.4 years] and ranged from 19 to 39 years. The patients were predominantly Caucasian (98.0%). Nearly one half of patients had no limitations due to malignancy at baseline, ECOG = 0 (49.0%). Married patients made up 42.6% of the population. Of young adults with sarcoma 28.6% had children (**Table 1**). Though the study allowed for patient data as far back as 1992, the earliest patient date of diagnosis was 2008.

Distribution by stage of cancer varied widely with 31.8% having stage I, 29.5% having stage II, 13.6% with stage III, and 25.0% with Stage IV disease per NCCN guidelines (Network Ncc. Nccn, 2021a,b,c). More YA patients had sarcoma of the extremity (57.1%%) than of the trunk (32.6%). The most frequently reported location of disease was the lower extremity 44.9% (**Table 1**).

Treatment

93.9% of patients underwent surgery within 1 year of diagnosis. Over one half (53.1%) of patients had limb surgery. Of those who had limb surgery 92.3% had limb-sparing surgery over amputation. 34.7% of patients underwent chemotherapy, hormone therapy, or targeted therapy and only 18.4% had radiation within the first year of diagnosis. All patients who underwent radiation in the first year received it as an adjuvant therapy (**Table 1**).

Patient reported quality of life by FACT-G

Self-reported FACT-G scores were recorded for 37 patients at baseline and 27 patients at 12 months (**Table 2**). The median time from diagnosis to completion of baseline questionnaire was 1 month and 12 months for the 1 year follow up survey. Differences between total eligible patients and self-reported FACT-G's existed due to various circumstances. Only

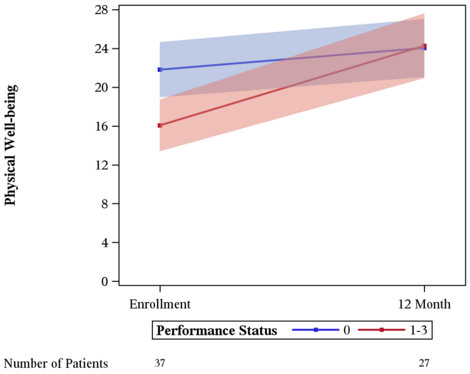

FIGURE 3
Changes in physical wellbeing by performance status.

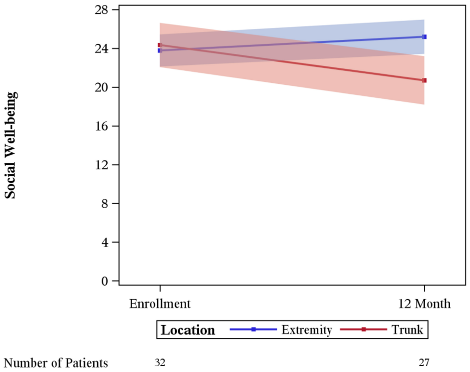

FIGURE 4
Changes in social wellbeing by anatomic location.

1 patient was lost to follow-up during this time period, resulting in 1 missing 12-month questionnaire. In addition, 1 baseline questionnaire was completed outside of the defined time window (within 6-months of the intended completion date). The remaining questionnaires at the respective time points are missing because patients did not return a completed questionnaire for unknown reasons.

Mean overall wellbeing scores also improved from baseline to 12 months (76.4 vs. 85.4, $p < 0.01$) (**Figure 1**). Physical wellbeing differed significantly between baseline reports and 12 months (18.8 vs. 23.9, $p < 0.01$) (**Figure 2**). Social wellbeing and emotional wellbeing did not differ significantly between baseline and 12 months; $p = 0.56$ and $p = 0.66$ respectively. Mean functional wellbeing scores were 16.5 at baseline and 19.8 at 12 months showing significant increase ($p < 0.01$) (**Figure 2**).

Significant differences in the rate of change between enrollment and 12-month FACT-G overall scores by age, sex, laterality, marital status, performance status, having children, clinical stage, limb surgery, chemotherapy, or tumor size were not evidenced. A statistically significant difference was seen for physical wellbeing among patients with no limitations (performance status of 0) vs. some limitation (performance status of 1–3) ($p = 0.03$), with those with some limitation showing a greater degree of improvement (**Figure 3**). A difference in social wellbeing scores was also seen based on location of malignancy, trunk vs. extremity ($p = 0.02$), with those with extremity showing greater improvement in scores (**Figure 4**).

Discussion

This retrospective review from an academic tertiary care center reveals that young adults with sarcoma report a perception of improved quality of life at 12 months after their diagnosis. Patients with decreased functional status at baseline report a higher change in quality of life, approaching those who did not have baseline limitations. Importantly, a difference is seen in changes in reported perception of social wellbeing depending on anatomical location with patients, with sarcomas of the trunk reporting decreased social wellbeing and patients with extremity sarcomas reporting improvements in social wellbeing. To our knowledge this analysis represents the largest study reviewing QoL outcomes in YA patients with sarcoma.

Overall FACT-G scores at enrollment were in line with non-GIST sarcoma scores reported in adults with a mean of 76.4 in this study and previous literature for adults with sarcoma reporting 76.4 and 75.49 (Ostacoli et al., 2014; Chan et al., 2015). Physical, Emotional, and Functional FACT-G scores were in line with previously reported study (Ostacoli et al., 2014). Social wellbeing in this YA cohort 24.4 were numerically higher than in cohorts of all adults 19.04 (Ostacoli et al., 2014).

When compared with childhood cancer survivors, physical wellbeing was numerically lower but with greater variability in the YA sarcoma patients in this study with a mean physical wellbeing of 18.8 in survivors of childhood cancer. In YA patients with sarcoma, baseline physical wellbeing scores are much lower than the general population with a mean of 25.1. social wellbeing scores seen in this YA sarcoma cohort are in line with AYA survivors, and higher than the United States general population mean 19.1 (Brucker et al., 2005; Bradford et al., 2021; **Figure 5**).

Physical wellbeing showed improvement in patients between their baseline and 1 year follow up. This could potentially be explained with treatment effect, and relief of pain, and limitations due to the location of their primary malignancy. Improvements in physical wellbeing scores were noted mostly in those who had decreased ECOG performance status at baseline. One could surmise that patients may have regained function and with treatment of their sarcoma noticed this improvement. In this study few patients underwent amputation and most underwent limb sparing procedures at a high-volume center which has been shown to be associated with improved overall survival (Abarca et al., 2018).

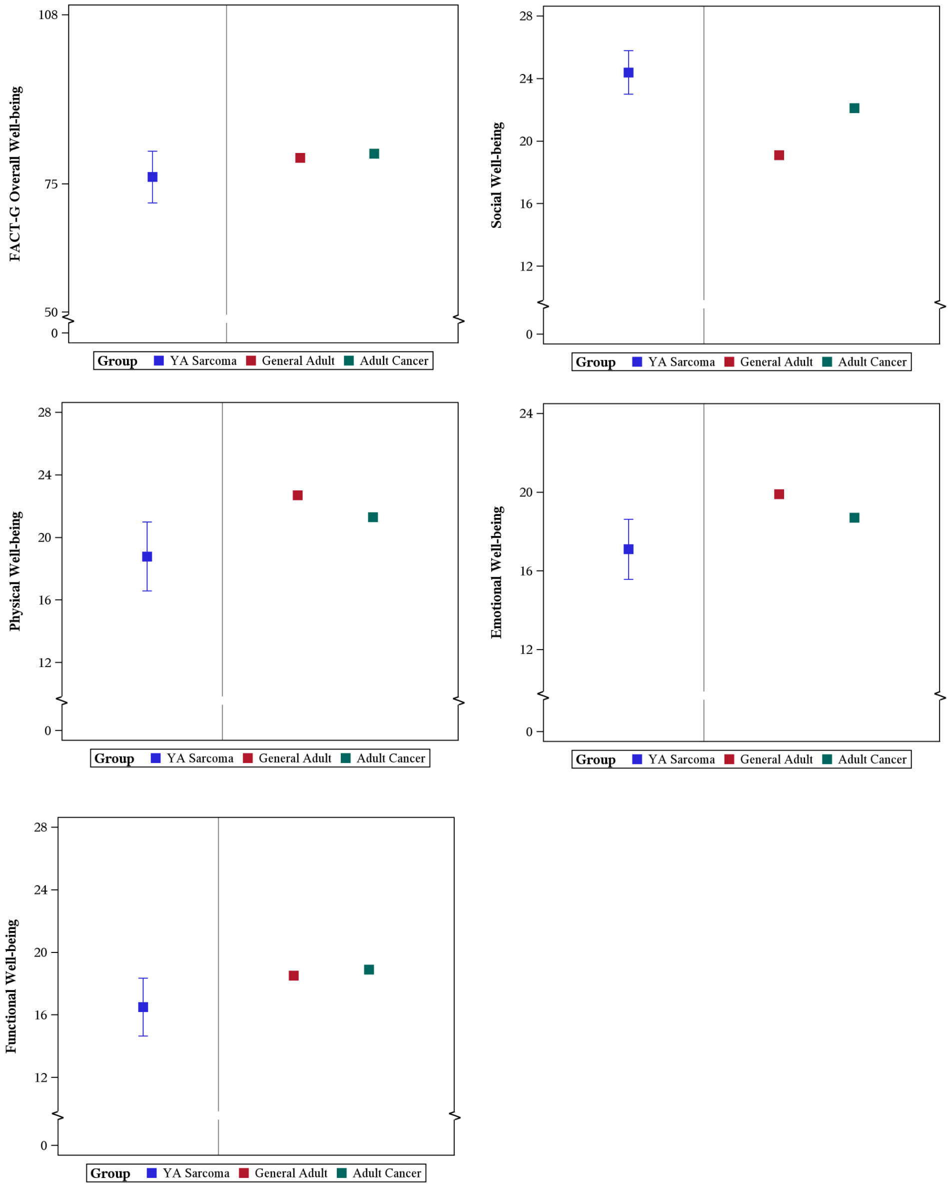

FIGURE 5
Comparison of YA patient reported FACT-G scores with General Adult and Adult with Cancer Population Scores.

Changes in social wellbeing had significant variation between patients with extremity vs. trunk sarcoma. This may relate to the improvements in wellbeing from being more active and able to be socially engaged after treatment of an extremity sarcoma. Limb sparing surgery which was done in >90% of the patients in this study may make this easier for patients with extremity sarcoma. Whereas patients with trunk sarcomas may require undergoing large surgical revisions and have significant scarring. This could be a source of embarrassment in some social situations. Other factors may depend on the extent of involvement of the sarcoma for example if there was bowel resection, or other organs were removed. Patients may have changes in intimate relationships or apprehension regarding sexual activity and body imaging post-surgical and chemotherapy treatments. Additionally, it is interesting that YA patients with sarcoma have numerically higher scores than age-matched peers or even age-matched peers with other malignancies (Brucker et al., 2005). This may be reflective of patients reaching out for support surrounding their diagnosis or is potentially reflective of the nature of the local culture.

For treating providers, clinical outcomes, such as overall survival and progression, seem paramount. One must not lose sight of other outcomes that matter to patients such as quality of life. Differences in patient-reported outcomes such as quality of life become important to track over time and should help serve as important endpoints for clinical trials in conjunction with subjective measures (Osoba, 2011). Ideally this will include longer follow up into patient survivorship to gain insight on the patient experience not just around initial treatment but how patients are cared for in the long term (Haslam et al., 2020).

This study has several important limitations that must be acknowledged. First this study is limited to the YA age group 18–39 and did not include adolescents; as patients ages 15–17 are not included in STiR at the University of Iowa. This study represents the experience of a single tertiary academic center and had a limited population that lacked diversity and may not be applicable to all patients. A limited sample size makes it difficult to understand the role of various factors in QoL. However, this limited data serves to aid in hypothesis generation for this understudied group. A response bias must also be acknowledged as some patients who are doing worse or are very ill may not have sent back surveys, possibly underestimating the true QoL in this population. Given that there is some variability for baseline enrollment and survey response there may be a concern of recall bias, this should be limited in that QoL is the only self-reported measure and all other data was derived from medical records. The patients that responded to the surveys may not be reflective of all patients.

Conclusion

Young adults with sarcoma treated at a tertiary center had improvements in overall reported QoL at 12 months. Overall baseline QoL scores on FACT-G were lower than the general population for YA patients with sarcoma but at 12 months were in line with general population norms. The improvements seen merit further investigation to evaluate how these change over the continuum of care and if interventions are needed at specific timepoints. Quality of life changes may be useful outcomes of interest in trials.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

This study was reviewed and approved by the University of Iowa Intuitional Review Board (IRB 202106171). Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

Author contributions

JRD, BL, SM, BM, and VM contributed to conception and design of the study. JRD, MC, BL, and SM organized the database. BL and SM performed the statistical analysis. JRD wrote the first draft of the manuscript. JRD, VM, BL, SM, and BM wrote sections of the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

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Conflict of interest

VM received research funding from Gateway Foundation, Rising Tide Cancer Research Foundation, Amgen. Advisory Board—Forma Therapeutics and Astex Pharmaceuticals. MM reports consultancy/advisory board for Blueprints Medicine, Immunocore, Amgen, Trieza, Array Biopharma, Biontech, and Novartis.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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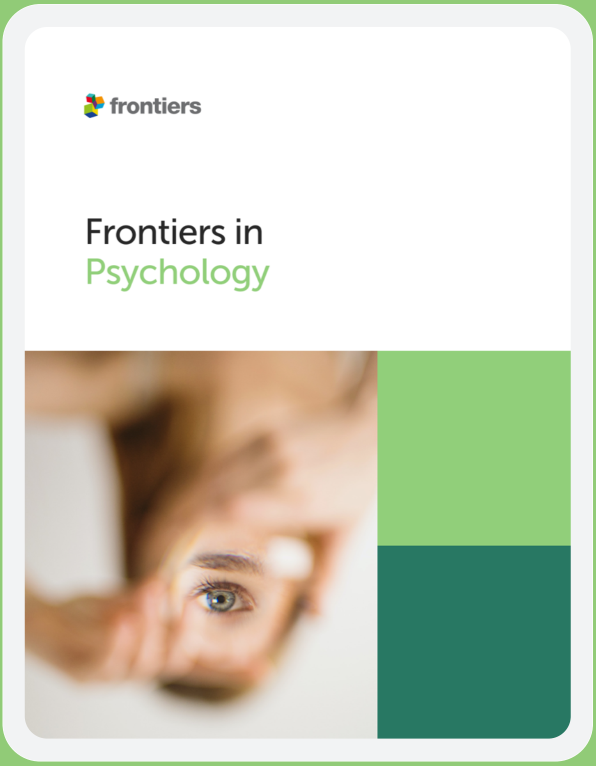

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